Understanding distress in women at increased risk of breast cancer. The effects of experience of breast cancer in the family and illness perceptions.

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DECLARATION

I hereby declare the work in this thesis to be my own, except where otherwise stated.

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DEDICATION

To Gran.

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LIST OF ABBREVIATIONS

BRCA1	Breast cancer susceptibility gene 1
BRCA2	Breast cancer susceptibility gene 2
BSE	Breast Self Examination
CBE	Clinical Breast Examination
CBT	Cognitive Behavioural Therapy
CFS	Chronic Fatigue Syndrome
CRC	Cancer Research Campaign
DCIS	Ductal Carcinoma In Situ
DNA	Deoxyribonucleic Acid
FAP	Familial Adenomatous Polyposis
FDR	First Degree Relative
GHQ	General Health Questionnaire
GP	General Practitioner
HBM	Health Belief Model
HIV	Human Immunodeficiency Virus
HRT	Hormone Replacement Therapy
IBIS	International Breast Cancer Intervention Study
ICRF	Imperial Cancer Research Fund
IMIQ	Implicit Models of Illness Questionnaire
IPQ	Illness Perception Questionnaire
IPQ-R	Illness Perception Questionnaire-Revised
LCIS	Lobular Carcinoma In Situ
MI	Myocardial Infarction
MRI	Magnetic Resonance Imaging
RA	Rheumatoid Arthritis
SRM	Self Regulatory Model
TRA	Theory of Reasoned Action
TPB	Theory of Planned Behaviour

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ABSTRACT

The aim of the current work was to improve understanding of factors associated with distress in women at increased risk of breast cancer because of their family history of the disease. Levels of distress have been described in this population although few studies have attempted to investigate causes of variation in distress. A number of anecdotal reports and qualitative studies have highlighted that women's experiences of breast cancer in their family is related to distress. The first study in this thesis aimed to assess women's experiences in a quantitative manner and demonstrated associations with general and cancer specific distress.

Applying theoretical perspectives from health psychology enables us to consider *how* these experiences may influence psychological response to genetic risk. Using Leventhal's Self Regulatory Model (SRM) (Leventhal et al. 1980) a theoretical model was developed for use in this thesis. This model proposed that perceptions of breast cancer mediate the impact of experiences of the disease in the family on psychological well-being. A large cross-sectional questionnaire study of women at increased risk of breast cancer and women in the general population was conducted in order to systematically explore this model. As there were no measures available to assess perceptions of breast cancer in healthy populations an existing generic measure was adapted and evaluated in the current samples. The mediation model was then systematically explored in a series of analyses.

The results confirmed a number of hypotheses. Women at increased risk of breast cancer showed higher levels of cancer specific distress and held different perceptions of breast cancer than women with no experience of the disease. Analysis indicated that experience of breast cancer in the family was associated with levels of distress and perceptions of the disease and that both experience of breast cancer and illness perceptions predicted distress in women at increased risk. Some support for the mediation model was found.

This thesis has shown that the SRM can be successfully applied to women at increased risk of breast cancer. Further work is required to explore additional aspects

of illness perceptions in healthy individuals at increased risk of disease and to test the causality of relations revealed in this thesis. Utilising theoretical models to understand response to risk in this clinical context is likely to provide implications for the development and evaluation of interventions aimed at improving psychological well-being.

OUTLINE OF THESIS

This section briefly outlines the overall organisation of the thesis. The research reported in this thesis aims to bring a theoretical understanding to a clinical issue-understanding variation in levels of distress in women at increased risk of breast cancer. The research is based on a large quantitative study designed to explore the impact of experience of breast cancer in the family and illness perceptions on levels of general and cancer specific distress in women at increased risk of breast cancer.

Chapter 1. begins with an overview of breast cancer. The risk factors, symptoms and prognosis of the disease are outlined as well as options for screening and treatment. This is followed by an explanation of genetic predisposition to familial breast cancer and the screening and preventative options available to women at increased risk. The psychosocial aspects of familial breast cancer are then explored with a particular focus on emotional response to breast cancer risk. Research that has examined the psychological well-being in women at increased risk of breast cancer is reviewed.

Chapter 2. focuses on the impact of experience of cancer in the family. The impact of women's experiences of breast cancer in the family on response to risk of familial breast cancer is reviewed. It is concluded that whilst this is an important determinant of response to risk research has tended to be descriptive in nature and a theoretical perspective is required to understand the mechanism involved.

Chapter 3. provides a discussion on theoretical perspectives that can be utilised to help understand and explain the relationship between experience of breast cancer in the family and response to risk. This chapter discusses the potential impact of personal experience on risk perception, decision making and illness representations. The Self Regulatory Model is described and a review of its application to understanding patient response to illness is outlined. The application of this model as a framework for understanding healthy individuals response to health threats is discussed.

Chapter 4. outlines the aims and design of the current research. The primary aim of the thesis is to test a mediation model that perceptions of breast cancer mediate the impact of experiences of the disease in the family on psychological wellbeing in women at increased risk of breast cancer. This is tested with a cross-sectional study including women at increased risk of breast cancer and a comparable sample of women in the general population. The objectives and hypotheses to be examined throughout the thesis are outlined and the design, participants, procedures and measures are described. Chapter 5. gives further details of samples included in the research and provides response rates and descriptions of samples. Chapter 6. is dedicated to describing in more detail the main measure utilised in the research. This chapter provides a detailed description of the development of a measure of experience and outlines pilot work to assess the acceptability and appropriateness of this measure for the current research. The adaptation and psychometric evaluation of an existing quantitative measure of illness perceptions for use in the current samples is also provided.

Chapters 7-10 comprise the results of the research. These chapters are organised to reflect different components of the theoretical model to be tested as outlined in Chapter 4. Chapter 7 provides the section of results referring to associations between experience of breast cancer and distress. Chapter 8. covers the analysis of associations between experience of breast cancer and illness perceptions. Chapter 9. reports the analysis of associations between illness perceptions and distress. Each of these chapters provides a comparison between the women at increased risk of breast cancer and a control sample as well as exploring associations within each sample. Chapters 7 and 9 also provide results of analysis to predict level of general and cancer specific distress in women at increased risk of breast cancer. Chapter 10. pulls these results together and systematically tests potential mediation models as highlighted throughout the subsequent results chapters.

Chapter 11 provides the main discussion for the thesis that builds on the summary discussions presented at the end of each chapter. Chapter 11. opens with a summary of the main findings presented throughout the thesis. This is followed by a discussion of the theoretical implications and methodological issues that have been raised in the

course of this research. This is followed by a discussion of the clinical application of this work and directions for future research and final conclusions.

CHAPTER 1

BREAST CANCER AND GENETIC PREDISPOSITION

BREAST CANCER AND GENETIC PREDISPOSITION

1.1 BREAST CANCER: CLINICAL AND PSYCHOSOCIAL ASPECTS

1.1.1 What is cancer?

Growth and development of cells in the body is regulated by complex genetic mechanisms that ensure tissues and organs develop, and remain, appropriate to the body's needs (Wienberg 1996). If the mechanisms controlling cell growth and division are disrupted, cells may divide in an uncontrolled manner causing the range of diseases collectively known as cancer. Cancer originates from one cell that has accumulated damage (or mutations) to the genetic controls over repeated cell divisions. It can occur in almost every tissue in the body and cancer cells are able to spread (metastasise) to other areas in the body. The series of genetic changes leading to cancer develop over a period of many years and both inherited and environmental factors are involved in this process. Accidents in cell division, failure of DNA repair mechanisms and environmental factors can all bring about genetic changes resulting in a cancer cell.

1.1.2 Breast cancer

Breast cancer is a malignant tumour that develops in the cells of the breast and is the most common form of cancer in women in the UK (excluding non-melanoma skin cancer) comprising 18% of all female cancers (McPherson 2000). In the UK, 26,000 new cases of breast cancer are diagnosed each year (Silva and Zurida 2000). Breast cancer is the leading cause of death in women aged 35-55 years (Baum and Schipper 1998). The average lifetime risk of breast cancer for women in the UK is 8% meaning that approximately 1 in 12 of all women in the UK will develop breast cancer in their lifetime. This figure represents cumulative risk for women who live to 85 years of age. About a third of all reported breast cancers occur in the older age group 70-85. The risk for women between the ages of 30 and 50 is 1/1000 per yr (2%) (Baum and Schipper 1998). Breast cancer can occur in men although it is very rare (Ravandi-Kashani and Hayes 1998).

1.1.3 Risk factors for breast cancer

There is no single cause of breast cancer and the disease is likely to be due to numerous interactions between genetic and environmental factors. A number of risk factors have been identified for breast cancer. These include family history; sociodemographic factors; exposure to radiation; age; previous benign disease; reproductive factors; exogenous hormones and lifestyle factors (McPherson et al. 2000). Although these risk factors are often widely publicised in the media the majority of risks are actually very low (with the exception of age, family history and previous atypical hyperplasia). A large proportion of women who develop breast cancer (about 66%) do not have *any* major risk factor (Harris et al. 1992). Baum and Schipper (1998) indicate that the evidence for risk factors is based on observational data which have *"methodological weaknesses and leading statisticians have questioned whether a relative risk below 2 is not within the experimental error of the method"*. (Baum and Schipper 1998, pg 13). There is little evidence to suggest that women can prevent breast cancer or reduce their risk by changes in lifestyle factors. Certain risk factors (ie oral contraceptives) need to be balanced with other risks such as unwanted pregnancy. The complexity concerning risk factors and different terms used to portray risk (ie absolute risk, relative risk and cumulative risk) may be difficult for the lay public to assess.

A number of models have been developed to calculate an individual's risk of breast cancer based on risk factors for the disease (McTiernan et al. 1997). The Gail model calculates breast cancer risk from epidemiological factors including age at menarche, age at first birth, number of affected relatives, number of previous breast biopsies and evidence of atypical hyperplasia (Gail et al. 1989). This model tends to underestimate risk due to family history because second-degree relatives and affected paternal family members are not recognised as increasing risk. The Claus model is based solely on family history information such as number of affected relatives and age of diagnosis (Claus et al. 1991). Neither model is able to provide a perfect risk estimate and reliability of the models over different populations has been questioned (Emery et al. 2000).

1.1.4 Symptoms and prognosis

Changes in breast cancer cells have been identified which allow the classification of pre-cancerous and cancerous cells. Minor changes in cells leading to cysts are known as hyperplasia or proliferative disease and may be due to either genetic changes in cells or hormonal changes within the cells' environment (Kelly 2000). Changes in the DNA of breast cells can result in non-malignant abnormal growth patterns known as atypical hyperplasia. Breast cancer shows greater DNA changes and is classified by the origins of the cancer (ie in ducts or lobules in the breast) and whether they are confined to the original site (in situ) or have invaded surrounding areas (invasive) (Baum and Schipper 1998). Further classification of invasive breast cancer also exists based on pattern of growth and cell morphology (Sainsbury et al. 2000). Controversy exists concerning the classification of ductal carcinoma and lobular carcinoma in situ (DCIS and LCIS) (Kelly 2000). These cells are unable to metastasise and hence may also be referred to as 'pre-cancerous' cells although they account for a large proportion of breast cancer diagnoses (Kelly 2000). Breast cancer cells do not necessarily follow this sequence of genetic changes and breast carcinoma in situ may not always lead to invasive disease. Post-mortem studies have revealed that progression from carcinoma in situ to invasive disease may be as low as 20% (Baum and Schipper 1998).

The number and type of symptoms associated with breast cancer varies widely depending on the stage of disease (how much it has spread) and the location of metastases. Pre-invasive carcinomas (DCIS and LCIS) have few symptoms and are often not detected until mammography. Invasive breast cancer can usually be felt as a hard and irregular lump. Changes in the skin, nipple and nipple discharge are also common, although pain in the breast is seldom a symptom of breast cancer (Baum and Schipper 1998).

Locally advanced breast cancer patients may also suffer from lymphoedema leading to swelling and possible pain and paralysis of the arm. Thickening and ulceration of the skin may also occur at the site of the tumour. Metastatic breast cancer (i.e. where cancer has spread from the affected breast) can cause a variety of symptoms depending on where the metastases occur. Common sites of spread are bone, liver

and brain resulting in a range of symptoms: pain, weakness, nausea, anorexia, weight loss, jaundice, headaches and neurological symptoms. The progression of breast cancer is extremely variable and the terminal phase of the disease may range from a few days to several months in duration. Baum and Schipper (1998) highlight the unpredictable nature of breast cancer:

> *"At one extreme, women may present with massive involvement of the axillary nodes or even bone marrow infiltration with the primary tumour virtually undetectable and die of breast cancer before the primary disease is clinically apparent. At the other extreme women may refuse treatment and live for 20-30 years with a slowly progressive cancer which though it may present an unpleasant problem for the patient, seems to lack the capacity to metastasise and kill"* (Baum and Schipper 1998 pg 21-22)

1.1.5 Screening

Primary prevention of breast cancer is not possible to date because of the lack of knowledge concerning pathogenesis of the disease. Screening aims to reduce mortality from breast cancer by detecting and treating the disease early before it has spread. Prognosis and survival from breast cancer depends on tumour size, stage of disease and metastases at presentation. Small tumours (<2cm) have a 5-year survival rate of more than 90%, compared to 60% for patients with tumours over 5cm (CRC factsheet 1996). Early detection is therefore paramount in reducing mortality.

Mammography is a screening test for breast cancer that uses X rays of the breast to detect abnormalities that may be malignant. Randomised controlled trials of mammography have shown that attendees benefit from reduction of up to 40% in breast cancer mortality with the highest reduction in risk shown in the 50-70 age group (Blamey et al. 2000). Cancers detected by mammography are likely to be smaller, non-invasive and less likely to have metastasised compared to symptomatic cancer (Blamey et al. 2000). The National Breast Cancer Screening programme in the UK offers mammography screening every 3 years to women in the age range 50-64 (Scottish Intercollegiate Guideline Network 1998).

The value of screening for younger women remains controversial. Analysis of data from randomised trials has shown no significant reduction in mortality for women

under the age of 50 (Fletcher et al. 1993, Kerlikowske et al. 1993). This may be because younger women's breasts are more dense making the test less sensitive. Breast cancer is also less prevalent in younger women reducing the number of cancers detected and cost effectiveness of screening in this group (Neugut and Jacobsen 1995). However there are concerns that screening techniques have been updated since these early trials. More recent studies have indicated that screening may be effective in reducing mortality from breast cancer in women as young as 40 (Tabar et al. 2001). The benefits of screening prove difficult to evaluate due to methodological problems. Large samples with long follow up periods are required in order to assess the impact of screening procedures on outcomes such as mortality. However this time span is confounded with changes in incidence and treatment over time (Wardle and Pope 1992).

There are a number of problems associated with screening. Firstly, the false negative rate is high (10-30%) as are the detection of unimportant abnormalities and false positives (approximately 1%) (Silva and Zurida 2000). A number of factors contribute to this including dense breast tissue, difficulties screening the entire breast, difficulties detecting lobular carcinoma and interpretation error (Silva and Zurida 2000). Adherence to mammography is not ideal and has been estimated at 70% (Baum and Schipper 1998). The optimal interval between mammograms has also not been determined. There are concerns that the time interval between screening is too long and that women reassured by screening procedures will not self examine between tests (Baum and Schipper 1998). Seventeen percent of tumours have been reported to occur between screening sessions (Andersson et al. 1988) and the majority of breast cancers are first detected by women themselves (Austoker 1994). Regular breast self-examination (BSE) is encouraged in order to increase early detection. Breast awareness in the UK, encourages women to become familiar with their breasts and to distinguish between normal cyclic fluctuations and abnormal changes. However there is no evidence that clinical breast examination (CBE) or BSE increase early detection or survival (Blamey et al. 2000) and no differences in mortality between those receiving BSE training and controls has been reported (Baum and Schipper 1998).

1.1.6 Diagnosis and treatment

Following identification of a breast lump subsequent investigations may include diagnostic mammography to identify the size and character of the abnormality, clinical examination and ultrasound. A tissue biopsy removing a sample of breast tissue may be conducted in order to determine if the lump is benign or malignant. Triple assessment (clinical examination, imaging investigations and pathological evaluations) are advised in order to gain a confident diagnosis (Scottish Intercollegiate Guidelines Network 1998, Baum and Schipper 1998). About 2/3 of breast abnormalities detected by mammography prove to be benign on further examination (Blamey et al. 2000).

Treatment for breast cancer may be targeted locally at the breast (surgery and radiotherapy) and systemically to treat metastatic disease. Local surgical treatments include mastectomy, in which the whole breast is removed with varying degrees of chest muscle and lymph glands, or breast conservation surgery to remove the tumour and surrounding tissue. Breast conservation surgery includes lumpectomy or quadrantectomy (removing a quadrant of the breast). The decision regarding the extent of surgery depends on several factors including the age of the patient, size of tumour relative to breast size, stage of tumour, grade of tumour (how fast the cancer cells are growing), patients preference and fitness for surgery and/or radiotherapy (Scottish Intercollegiate Guidelines Network 1998). Mastectomy is recommended for approximately 1/3 of localised disease (Sainsbury et al. 2000). There are a number of possible complications following mastectomy including bruising, swelling, infection, nerve damage, shoulder weakness or stiffness and swelling of arm due to lymphoedema. Radiotherapy targets high energy rays to kill cancer cells and is used after surgery to prevent local recurrence or metastases. It is normally administered in daily sessions with breaks at weekends for up to 6 weeks. Common side effects include skin reactions, nausea and vomiting. Pneumonitis from irradiation of the lungs is a rare side effect occurring in less than 2% of patients (Baum and Schipper 1998).

Although breast cancer originates in the breast it may spread to other parts of the body. Systemic treatments target cancer cells anywhere in the body and include

chemotherapy and hormonal treatment. These treatments may be delivered before surgery to reduce size of lump and/or following surgery to reduce risk of spreading (adjuvant therapy). Often patients with distant metastases are incurable and in these cases systemic treatment is provided in order to relieve symptoms and maintain quality of life.

Chemotherapy can be given as tablets or intravenously and a full course can take up to 6 months to complete. Side effects of chemotherapy will vary according to the specific drugs used but commonly include tiredness, hair loss, nausea, risk of infection, diarrhoea, vomiting, risk of infertility, premature menopause and weight gain. Hormone therapy uses drugs such as tamoxifen that reduce the impact of oestrogen on cancer cells. These drugs are taken over a period of at least 5 years following surgery (Scottish Intercollegiate Guidelines Network 1998) and side effects include menopausal symptoms such as hot flushes and slight increased risk of endometrial cancer. Adjuvant systemic therapy using chemotherapy and/or hormone therapy has been shown to reduce the incidence of recurrence of breast cancer and overall mortality (Early Breast Cancer Trialists' Collaborative Group 1988, 1992). Tamoxifen has been shown to reduce the chance of recurrence by 25% and mortality by 17% although the benefits are largely selective to patients with oestrogen receptor positive cancers (Early Breast Cancer Trialists' Collaborative Group 1992, 1998). Ovarian ablation is another possible treatment that has been found to reduce recurrence and death from breast cancer in pre-menopausal women with oestrogen receptor positive cancers (Early Breast Cancer Trialists' Collaborative Group 1996). Side effects from this procedure include early menopause and infertility.

A number of problems have been identified in women subsequent to treatment for breast cancer that may affect quality of life. These include lymphoedema due to damage to the lymph nodes following treatment, problems with arm mobility, menopausal symptoms and cosmetic issues. Women who have been treated for breast cancer are also at high risk of clinical levels of anxiety and or depression, sexual difficulties and body image problems (Maguire et al. 1978, Irvine et al. 1991).

1.1.7 Recurrence

Following treatment patients are maintained under regular surveillance in order to check for signs of recurrence and to ensure metastases are detected as early as possible. The effectiveness of mammography following breast conservative surgery is reduced since scarring resembles the appearance of cancer (Sainsbury et al. 2000). Recurrence has been associated with disease stage at diagnosis and extent of surgery (more extensive surgery is associated with lower recurrence). Recurrence following breast conservation surgery and radiotherapy is at the frequency of about 1% per annum (Scottish Intercollegiate Guidelines Network 1998). Local recurrence of cancer following mastectomy is most likely to occur within 2 years following surgery (Sainsbury et al. 2000) and is often associated with distant metastases within the subsequent decade (Scottish Intercollegiate Guidelines Network 1998).

Mortality from breast cancer has been declining since 1990 in America and Europe (Mettlin 1999). This decline is likely to be due to earlier diagnosis with mammography and more effective adjuvant therapy. Trials are currently being conducted to test the best combinations of treatments, timing, doses and methods of delivery as well as use of new drugs on survival and quality of life (Baum and Schipper 1998). There are a number of new treatment possibilities that are currently being tested including vaccines which trigger the bodies immune response and treatment aimed at triggering the cancer cells suicide gene (Imperial Cancer Research Fund- Cancer Information Service). Although no preventative measures are available at present a number of potential areas are under investigation for example use of tamoxifen as a preventative agent.

1.1.8 Summary

Breast cancer is one of the most common forms of cancer although medical understanding of the pathogenesis of the disease is unclear. A number of risk factors have been associated with the disease although the majority are uncontrollable and there is little evidence to suggest that changes in lifestyle factors can prevent the occurrence of the disease. Breast cancer has few observable symptoms although may be detected by changes in the breast. Screening with mammography in older women is effective in reducing mortality although remains controversial in women under the

age of 50. The false negative rate in mammography screening is high and there are concerns regarding the optimal timing of screening sessions. Both CBE and BSE are of unproven efficacy across all age groups to date. Women in the general population therefore face a fairly high risk of breast cancer with little scope for personal control and the detection methods available are controversial and anxiety invoking. The prognosis of breast cancer is extremely variable and treatment side effects may be severe. Psychosocial problems following breast cancer are common and patients face a risk of recurrence.

1.2 FAMILIAL BREAST CANCER: CLINICAL ASPECTS

1.2.1 Genetic predisposition to breast cancer

A family history of breast cancer has long been established as a risk factor for the disease. Epidemiological studies have shown that individuals with affected relatives have a higher risk of breast cancer and are more likely to develop the disease at a younger age (Adami et al. 1980). It has been estimated that about 5-10% of breast cancer cases are caused by hereditary factors. Studies of families with high incidence of breast cancer suggested that the disease showed patterns indicative of dominant inheritance (Claus et al. 1990). Molecular genetic studies have been conducted to localise the genes linked with breast cancer and estimate their penetrance (Hall et al. 1990, Easton et al. 1993). In 1994 the BRCA1 gene associated with both early onset familial breast and ovarian cancer was identified on chromosome 17 (Miki et al. 1994, Futreal et al. 1994) followed by discovery of a second gene (BRCA2) on chromosome 13 (Wooster et al. 1995).

BRCA1 and BRCA2 are large genes that encode proteins involved in DNA repair (tumour suppressor genes). Mutations can occur at any location on these genes and are associated with a significant increased risk of developing breast cancer. Over 100 mutations on the BRCA1 gene have been identified (Collins 1996). The BRCA1 and BRCA2 genes are autosomal dominant so the offspring of a mutation carrier has a 50% chance of inheriting the gene. Mutations can be inherited from either the maternal or paternal side of the family. Hereditary breast cancer occurs at a younger age because these germline genetic mutations are present in all cells at conception. It

therefore takes less time for additional mutations to occur and subsequently for cancer to develop compared to non-hereditary breast cancer caused by accumulated genetic mutations in somatic cells throughout an individual's lifetime. Most breast cancers that are due to a genetic mutation occur before the age of 65. Mutations in either BRCA1 and BRCA2 are implicated in up to 25% of patients diagnosed with breast cancer before age 40 (Baum and Schipper 1998).

BRCA1 and BRCA2 show high but not full penetrance. Lifetime risks associated with a mutation in either gene were initially estimated at about 80%. These penetrance rates are likely to be overestimated because they are based on studies of families with extremely high incidence of breast cancer (Collins 1996). More recent studies of mutation carriers have shown lower penetrance estimates of under 60% (Struewing et al. 1997). The risk of developing breast cancer is therefore uncertain even for known mutation carriers. BRCA1 mutations are also associated with an increased risk of ovarian cancer and male carriers are at an increased risk of developing colon cancer and prostate cancer (Silva and Zurrida 2000). The risk of ovarian cancer is not as high for BRCA2 carriers although mutations on this gene are also associated with an increased risk of a range of other cancers including male breast cancer, cancer of the uterus, prostate cancer, pancreatic cancer and gastric cancer. Breast cancer patients carrying BRCA1 or BRCA2 mutations are at high risk of contralateral breast cancer.

BRCA1 and BRCA2 are implicated in approximately 4% of breast cancers (Baum and Schipper 1998) and are currently only thought to account for families with an extremely strong family history (ie at least 5 cases of breast cancer under the age of 45 or ovarian cancer at any age) (Emery et al. 2000). However, there are also likely to be other more common mutations with lower penetrance associated with a moderate increased risk of breast cancer (Ford et al. 1995).

The role of environmental factors in phenotypic expression of risk is unclear. Although studies have been carried out to determine interactions between family history and other risk factors including reproductive factors and exogenous hormones the results are controversial (Evans et al. 1994). In a meta-analysis of studies of HRT

use and breast cancer risk Steinberg et al. (1991) found that use of HRT significantly increased the risk of breast cancer for women with a family history. Dupont and Page (1985) also found the risk associated with proliferative breast disease was significantly greater in women with a family history compared to those without. A major problem with many studies of this type is the definition of family history. Many studies define 'family history' as at least one first degree relative with breast cancer although many such cases may not actually represent a significant family history of the disease. This reduces the ability to detect effects within true familial cases (Evans et al. 1994).

1.2.2 Risk assessment

Media interest in breast cancer and publicity concerning BRCA1 and BRCA2 has lead to increased awareness of genetic causes of breast cancer. Cancer genetic services have developed in order to provide information and advice about familial cancer, to identify individuals at increased risk of cancer and to establish effective management strategies (The Scottish Office 1998). Women are mainly referred to familial breast cancer clinics from primary care and studies have indicated a large increase in GP consultations regarding family history of cancer and referrals to cancer genetic clinics (Emery et al. 2000; The Scottish Office 1998). Although approximately 8% of women over 40 have at least one first degree relative with breast cancer only a small proportion of these women will have a significant family history of the disease (Mettlin 1994). Familial breast cancer clinics therefore conduct a detailed analysis of family history in order to obtain a genetic risk estimate. Assessment of family history is prone to a number of problems. For example, little is known about the accuracy of self reported family histories and errors may result from lack of knowledge and accuracy of previous diagnoses and/or medical records (Richards 1999). Families may also not include a sufficient number of females to allow expression of the condition (Richards 1999).

Current genetic services have developed due to local pressure in a 'fragmented and uncoordinated manner' (The Scottish Office 1998). As a result there is as yet no UK national consensus for determining risk status and guidelines for assessing family history vary in procedure and recommendations (Emery et al. 2000). The criteria

provided by the National Clinical Guideline for Scotland are outlined in Figure 1.1 (Scottish Intercollegiate Guideline Network 1998). The crucial aspects of family history include: the age at which affected relatives were diagnosed; the number of relatives diagnosed; their relation to the counsellee and the site of their cancer (unilateral or bilateral breast cancer and incidence of other cancers). Young age of onset of breast cancer is considered more important than the number of affected relatives in the family (Silva and Zurrida 2000). The empiric risk of familial breast cancer for an individual is not static but changes with events in the family (such as diagnoses) and age of counsellee. Women with a positive family history are at greatest risk between the ages of 30-50 years. As their age increases beyond this level without occurrence of breast cancer the chances of having inherited a mutation and developing familial breast cancer are reduced (Evans et al. 1994). However their risk falls to the level of the general population risk of sporadic breast cancer and this risk increases with age.

Figure 1.1- Criteria for identifying women at substantial increased risk

The following categories identify women who have three or more times the population risk of developing breast cancer.

A woman who has:

- One first degree relative with bilateral breast cancer or breast and ovarian cancer **or**
- One first degree relative with breast cancer diagnosed under the age of 40 years or one first degree male relative with breast cancer diagnosed at any age **or**
- Two first degree or second degree relatives with breast cancer diagnosed under the age of 60 years or ovarian cancer at any age on the same side of the family **or**
- Three first degree or second degree relatives with breast and/or ovarian cancer on the same side of the family

In this context a first degree relative is mother, sister or daughter. A second degree female relative is grandmother, granddaughter, aunt or niece.

Criteria for identifying women at very high risk in whom direct gene testing might be appropriate:

- Families with four or more relatives affected with either breast or ovarian cancer in three generations and one alive affected individual.

1.2.3 Screening and detection

An increased understanding of genetic predisposition to breast cancer has the potential to reduce mortality by enabling surveillance programmes and prevention trials to be targeted to those at increased risk. In Scotland women who are deemed at significant increased risk of breast cancer because of their family history are offered regular clinical examination and mammography. It is recommended that screening starts at 35 or 5 years younger than the youngest affected relative, whichever is first. Women under 40 are offered biennial mammography and annual clinical examination, women aged 40-50 are offered annual mammography and clinical examination. Women aged over 50 are either discharged to the national screening programme or continue with annual screening (Scottish Intercollegiate Guideline

Network 1998). Although guidelines for risk management are under development (Vasen et al. 1998, Eccles et al. 2000) there are currently no internationally agreed protocols to determine when strategies such as screening and genetic testing should be offered.

The effectiveness of screening among younger women in the general population remains controversial but data are emerging to support its effectiveness among younger women selected for their familial risk (Eccles et al. 2000, Macmillan 2000, Lalloo et al. 1998, Kollias et al. 1998, Tilanus-Linthorst et al. 2000). The detection rate ratio of benign and malignant biopsies has been found to be comparable to the National screening programme. In addition a higher proportion of in situ cancers are identified compared to a symptomatic sample matched for age and family history (Lollias et al. 1998, Lalloo et al. 1998). Although the significance of detecting non-invasive tumours is unknown it is estimated that 30-50% of these lesions will become invasive. Tilanus-Linthorst et al. (2000) compared annual surveillance of women at increased risk with symptomatic presentation of women with a family history and found significantly more cancers were detected at an earlier stage in the screened sample. This may suggest the need for more frequent screening in younger women to detect breast cancer at an early stage given the aggressive nature and rapid development of early onset breast cancer (Neugut and Jacobsen 1995). However shortening the screening interval holds cost implications. Although these studies are optimistic they are of limited sample size and duration. No randomised trials to explore the effectiveness of screening younger women with a family history have been conducted and the impact of screening on mortality is not known in this population. Both the clinical and cost effectiveness of these services remains undetermined (The Scottish Office 1998). Trials are currently in progress to assess new screening methods such as magnetic resonance imaging (MRI). Initial trials of MRI screening for breast cancer reported higher sensitivity and specificity compared to mammography (Kuhl et al. 2000).

1.2.4 Genetic testing

Women at high risk of breast cancer may be offered the possibility of genetic testing for a mutation in their family. The first stage of genetic testing involves identification

of a mutation in an affected relative ('diagnostic testing'). Both the BRCA1 and BRCA2 genes are large and mutations can occur at any position making diagnostic testing demanding and time-consuming. It is essential therefore that genetic testing is offered only to women with a strong family history suggestive of autosomal inheritance. If a mutation is identified 'predictive testing' can be carried out on a-symptomatic individuals to determine if they have inherited the specific mutation. The utility of predictive testing for genetic predisposition to breast cancer has been criticised due to the uncertainty of penetrance estimates and limited options for surveillance and prevention strategies available to mutation carriers (Evans et al. 2001). Testing may also be non informative since not every possible mutation will be detected and there may be other moderate risk genes that remain to be discovered

1.2.5 Prevention

There are no strategies of proven effectiveness for preventing breast cancer in those who test positive. Preventative options that may be considered include chemoprevention or prophylactic surgery. Tamoxifen was considered for prevention of breast cancer because it was shown to be effective in reducing risk of relapse of breast cancer in affected patients. A number of randomised controlled studies have assessed the impact of tamoxifen on risk reduction, although the results are inconsistent. A large randomised placebo controlled trial of over 13,000 women in the US reported a large reduction in risk of breast cancer (Fisher et al. 1998). The sample consisted of women at increased risk of breast cancer for a variety of reasons: their age (60 years or older); a history of LCIS or an increased risk of breast cancer determined by the Gail model (see 1.1.3, page 18). It was reported that tamoxifen reduced the risk of invasive cancer by 49% and non invasive cancer by 50% across the sample. The reduction in risk was found to be true across all subgroups although the effect appeared to be particular to oestrogen positive tumours. Side effects reported from tamoxifen use included increased risk of endometrial cancer, stroke, pulmonary embolism and deep vein thrombosis.

Two further randomised placebo controlled trials of tamoxifen in the UK (Powles et al. 1998) and Italy (Veronesi et al. 1998) found no effect of tamoxifen on breast cancer prevention. However a number of differences in methodology and samples

may account for differences in results. The Italian study was based on a sample of women who had had a hysterectomy and were at lower risk of breast cancer than age matched controls who had not had hysterectomy. The study also included a high proportion of women using HRT (Veronesi et al. 1999). The statistical power of the study was also low due to a small sample size and low compliance rate. The UK study was based on nearly 2500 women with a family history of breast cancer (Powles et al. 1998). Despite a high compliance rate and sufficient statistical power to detect differences between the groups no difference in frequency of breast cancer was found between the experimental and control samples. The sample in this study was women at familial risk of breast cancer compared to the US study whose sample was predominately based on non-genetic risk factors. This raises the possibility that oestrogen may be less important in the development of familial breast cancer reducing the effect of tamoxifen in this group. Further assessment of the effects of tamoxifen in women with a family history needs to be conducted with larger sample size and longer follow up. A European double blind randomised trial (International Breast Cancer Intervention Study- IBIS) is currently underway to determine the benefits and risks associated with long term tamoxifen use in healthy premenopausal women at increased risk.

Women at high risk who have tested positive for a genetic mutation may consider prophylactic bilateral mastectomy in order to reduce their risk of developing breast cancer. A retrospective study of women undergoing prophylactic mastectomy found a 90% reduction in incidence of breast cancer (Hartman et al. 1999). However no epidemiological data or controlled prospective studies are available to prove the effectiveness of the procedure and a residual risk of breast cancer still remains.

BRCA1 mutation carriers also have to contend with managing an increased risk of ovarian cancer. Currently recommended screening techniques include tranvaginal ultrasound and assessment of CA125 levels. Potential chemoprevention includes use of oral contraceptives which has been shown to reduce the risk of ovarian cancer in the general population by up to 60% (Berchuck et al. 1999). Prophylactic oophorectomy may also be considered although the benefits and side effects of this

procedure are undetermined (Berchuck et al. 1999, Fry et al. 2001). Evidence for all these strategies is limited and clinical trials are required (Emery et al. 2000).

1.2.6 Treatment

Women with a family history of breast cancer who develop breast cancer with a good prognosis may be advised to have a bilateral mastectomy because of the risk of contralateral breast cancer (Scottish Intercollegiate Guidelines Network 1998).

Breast cancers associated with BRCA1 and BRCA2 mutations have shown different cellular pathology compared to sporadic cases (Lakhani et al. 1997). This raises the possibility that different treatment strategies may be recommended. Further clinical research is needed to address these issues.

1.2.7 Summary

A family history of breast cancer has been recognised as a risk factor for the disease for many years. Recent research has identified autosomal dominant genes that when mutated predispose the carrier to developing breast cancer and are also associated with heightened risk of other cancers. These genes do not show 100% penetrance and the role of environmental factors is unclear. Although many women will have a relative who has suffered from breast cancer a small proportion are likely to be at increased risk of the disease. Familial breast cancer clinics have been established in order to provide risk assessment and also screening for those women who are deemed to have a significant family history. Although genetic testing is widely publicised it is technically difficult and not available to the majority of attendees. There are no proven preventative options for breast cancer. Chemoprevention and prophylactic surgery are only offered within the context of a clinical trial and may produce distressing and debilitating side effects. Women with a family history of breast cancer therefore face much uncertainty regarding if, when and where cancer will develop and how they should manage their risk.

1.3 FAMILIAL BREAST CANCER: PSYCHOSOCIAL ASPECTS

1.3.1 Psychosocial issues

Although knowledge concerning cancer genetics has increased within the past decade understanding of how to apply this information in clinical practice without adverse consequences for the individuals concerned, is limited. As the above review has highlighted *"advances in the basic sciences may occur at a pace that outstrips health services and genetic epidemiological research into the clinical implications and applications of these discoveries*" (Emery et al. 2000, pg 11). Given the complexity of information regarding breast cancer risk, the uncertainty of risk estimates and unproven efficacy of management options there are likely to be a number of psychosocial consequences of cancer risk for the individual and their families.

Research has begun to address the range of psychosocial issues surrounding familial cancer risk (Hopwood 1997). These issues include the process and outcomes of cancer risk counselling, decision-making, psychosocial effects of surveillance and genetic testing as well as longer-term adjustment to risk. The following section of this chapter will review the current work on psychosocial issues in breast cancer genetic risk. Firstly, characteristics of women attending for breast cancer risk counselling will be described and the difficulties of communicating cancer risk outlined. Secondly, the cognitive, behavioural and emotional outcomes of genetic predisposition to breast cancer will be discussed.

1.3.2 Women attending familial breast cancer clinics

The vast majority of women attending familial breast cancer clinics in the UK are Caucasian and well educated with a mean age of around 40 (Cull et al. 1999, Watson et al. 1999, Brain et al. 2000). Women from ethnic minorities and those with limited education are under represented at familial breast cancer clinics, as are women with a paternal as opposed to maternal family history (Richards 1999). The majority of women are referred to the centre by their GP following a discussion about family history of breast cancer initiated by the counsellee. The main reasons given for attending the clinics include: to obtain information about personal risk and risk status

of other family members, because of awareness of family history, to reduce anxiety, to obtain genetic testing for access to breast screening and information about prevention (Brain et al. 2000).

Earlier studies in the US attempted to describe women at increased risk in terms of their risk perception and levels of distress. Kash et al. (1992) assessed women at high risk of breast cancer because of their family history who were attending for screening at a familial centre. Twenty-seven percent of these women were found to show levels of distress that warranted clinical investigation. Seventy-six percent of women believed they were at moderate or high risk of developing breast cancer. Lerman et al. (1994a) identified a sample of nearly 800 women at increased risk of breast cancer either through an affected relative or women who had self referred to a prevention-screening centre. Over two thirds of the sample perceived their breast cancer risk as high and levels of distress in this population were higher than the general population (although depression and mood disturbances were comparable). Neither study provided information about the accuracy of women's risk perception relative to their actual risk status. Accuracy of risk perceptions and levels of distress prior to genetic counselling have been explored in UK studies. Cull et al. (1999) assessed women attending the familial breast cancer clinic in Edinburgh and found that the majority of women held fairly accurate risk perceptions. Only 14% overestimated their risk compared to 39% of women who underestimated their risk. However, a small proportion of women (n= 18, 4%) believed it was inevitable that they would develop breast cancer. Mean trait anxiety in the sample was higher compared to women in the general population of the same age but comparable to that found in older women attending routine mammography screening. Watson et al. (1999) assessed 303 first time attendees with a family history at a familial breast cancer clinic in London. Fifty-two percent of women were found to overestimate their risk and 18% underestimated their risk prior to counselling. A third of participants showed notable levels of distress although anxiety was similar to screening populations.

1.3.3 Communicating risk

The aim of genetic counselling is to collect and analyse medical and family history information in order to provide information regarding personal risk status and education about measures available for cancer control and prevention. Women need to comprehend a number of risk figures including the likelihood that the cancer in the family is due to an inherited mutation, the likelihood of carrying mutation, the probability of developing cancer and the chance of passing a mutation on to offspring. It is important that individuals understand these risk concepts to equip them to make an informed choice about their subsequent health management. Perception of the probability of developing cancer has widely been used as a main outcome measure by which to evaluate the effectiveness of genetic counselling. However, studies assessing the impact of genetic counselling on risk perception have been inconsistent. Some studies have shown improvements in accuracy or risk perception subsequent to genetic risk counselling (Evans et al. 1994, Gagnon et al. 1996) whereas other studies suggest inaccuracies are maintained (Lerman et al. 1995a). A number of follow up studies of genetic counselling have shown that although some women modify their risk perception towards the counselled risk a significant proportion of women continue to overestimate or underestimate their risk following genetic counselling (Cull et al. 1999, Hopwood et al. 1998, Lloyd et al. 1996, Watson et al. 1999).

A large number of factors may influence interpretation and recall of risk estimates (Kelly 1992, Hallowell and Richards 1997). The manner in which information is expressed may influence interpretation. There are numerous ways to present risk information in both qualitative and quantitative formats (eg high/low risk, absolute risk, relative risk, cumulative life time risk, % increase in risk etc). In a review of studies assessing communication of risk information Bottorff et al. (1998) concluded that the different ways currently used to communicate cancer risks have not been adequately evaluated. There is no clinical consensus for how to best communicate risk information in order to maximise understanding and recall. Cognitive bias may also affect interpretation of such probabilistic information (Tversky and Kahneman 1974, Lippman-Hand and Fraser 1979, Kessler and Levine 1987). This been demonstrated within the genetic counselling situation (Shiloh 1994). Methodological

differences between studies in definition and communication of risk, time delay between counselling and risk assessment and provision of written information after counselling may account for some inconsistencies in the literature. There are no standard measures of risk perception and measures do not capture what the risk *means* for the individual.

1.3.4 Screening behaviour

In order to achieve health benefit, provision of genetic risk information must encourage participation and adherence to the recommended screening practices. Although women with a family history of breast cancer are more likely to report they have had mammographic screening than those without a family history (McCaul et al. 1996) research examining screening behaviour in women with a family history of breast cancer has shown variable adherence to mammography and CBE (Meiser et al. 2000). Adherence to BSE has been shown to be fairly poor (Meiser et al. 2000, Alagna et al. 1987). The vast majority of studies examining screening behaviour in women at increased risk have been conducted in the US where lower adherence rates may reflect the personal responsibility for cost of screening. This has been indicated by associations between mammography use with income and employment in US samples (Lerman et al. 1993).

Psychological distress has been associated with uptake of screening in women with a family history of breast cancer although studies show conflicting results. Lerman et al. (1993) reported a negative association between breast cancer specific distress and adherence to mammography and Kash et al. (1992) found a negative association between distress and adherence to BSE and CBE. Other studies have reported positive associations between distress and screening behaviour (eg Meiser et al. 2000). Lerman and Schwartz (1993) suggest that the relations are complex and may indicate a curvilinear relation between anxiety and screening. This is consistent with the fear arousing communication theory which proposes that moderate levels of anxiety motivate screening behaviour whereas high levels of distress results in avoidance (Janis and Feshbach 1953). Reviews of fear arousing communications however, have shown that fear is effective in producing behaviour change if individuals have high efficacy (Witte and Allen 2000, Sutton, 1982). This suggests

that women's beliefs about the efficacy of screening methods and self efficacy in performing BSE need to be considered when investigating associations between distress and screening behaviour.

Studies assessing screening use in women at increased risk of breast cancer have a number of limitations. The majority of studies are based on retrospective self reports of screening which have potential for bias associated with recall and social desirability. Although self reported mammography use has been shown to be valid when compared to objective records (Etzi et al. 1994), prospective studies are required to confirm retrospective results. Differences between study samples, in knowledge and awareness of risk, risk status, counselling procedure and awareness of screening guidelines, can make comparisons between studies difficult. Samples selected through an affected relative are not directly comparable to those with a strong family history of breast cancer and samples recruited from familial breast cancer clinics are often based on women who are likely to be highly motivated to adhere to screening and are generally of a high educational level. The generalisation of results beyond self selected samples of women at increased risk may therefore be problematic (Meiser et al. 2000).

Cross-sectional correlational studies linking anxiety and screening (eg Kash et al. 1992, Lerman et al. 1993) can also not rule out the possibility that engaging in screening causes anxiety. Detection behaviours in which individuals face the risk of cancer are likely to elicit greater emotional responses than prevention behaviours. A number of psychological costs of screening for cancer have been outlined by Wardle and Pope (1992) and include anxiety provoked by publicity about screening and invitations to attend, discomfort surrounding the practical procedure of the test, detection of abnormality and false positives, stress associated with waiting for result and suspecting cancer and trauma associated with diagnosis of cancer. In a qualitative study of women attending the Edinburgh familial breast cancer clinic for surveillance because of their increased risk of breast cancer women described how their anxiety fluctuated and increased during the month in which the screening test was expected, when the appointment letter was received, during the screening procedure and on receipt of results (Appleton et al. 2000). A number of quantitative studies using validated measures of anxiety have suggested that false positive results

are associated with temporary increased levels of anxiety but this effect is not maintained at long term follow up (Lerman and Rimer 1993, Fentiman 1998). Further research is needed in this area to determine psychological costs of screening women at increased risk of breast cancer and to identify vulnerable groups of women.

1.3.5 Genetic testing: Interest and uptake

Interest in genetic testing for BRCA1 and BRCA2 mutations has been reported to be as high as 90% in women with a first degree relative with breast cancer (Lerman et al. 1994b, 1995b). Reports of actual uptake of genetic testing are lower. Lerman et al. (1997) reported an uptake of 58% in 149 individuals from high risk families in the US and Watson et al. (1996) reported an uptake rate of 41% among 2 high risk families in the UK. Although the uptake rates are lower than would be predicted from level of interest in genetic testing for BRCA1 and BRCA2 the rates exceed that reported for other late onset genetic disorders such as Huntingtons Disease. This is likely to reflect the difference between the disorders in terms of penetrance rates and options for detection and prevention.

A number of studies have addressed reasons for requesting predictive testing for breast cancer. Important factors include: to obtain certainty about risk status (Kash et al. 1997, Lodder et al. 1999); to know the necessity for surveillance (Lerman et al. 1994b, 1995b, Kash et al. 1997, Lynch et al. 1999 Lodder et al. 1999); to consider prophylactic surgery (Lodder et al. 1999); and to know the risk status of children (Lerman et al. 1994b, 1995b, Lynch et al. 1999, Lodder et al. 1999). Lerman et al. (1996, 1997) found that actual uptake of predictive testing was positively associated with level of education, cancer specific distress, possessing health insurance and number of affected relatives. Women were more likely to undergo genetic testing than men and uptake was associated with more knowledge about genetic testing and greater perception of the importance of benefits of testing (Lerman et al. 1996). Jacobsen et al. (1997) looked at the decision making process of obtaining predictive testing for breast cancer in women with first degree relatives in the US. Likelihood of taking a test was greater for women who perceived more pros than cons of the test. Commonly cited benefits of testing included helping other relatives to make

decisions about testing, to motivate BSE, to decide about prophylactic surgery and to reduce concerns if tested negative. Disadvantages of testing included increased anxiety and concern about other relatives. Reasons for declining predictive testing have included fear of insurance discrimination and fear of a positive test result (Lynch et al. 1999, Lerman et al. 1995b). Uncertainty about when breast cancer might occur and concern over the accuracy of the test have also been noted as important factors associated with rejection of predictive testing (Kash et al. 1997, Lerman et al. 1995b).

1.3.6 Genetic testing: Experience and response

Lodder et al. (1999) looked at experiences of women waiting for BRCA1 and BRCA2 test results in Holland. The majority of women showed normal levels of anxiety, although a significant proportion (25%) showed high levels of general and cancer-specific distress. Higher levels of distress were found for: women who were anticipating problems and were considering prophylactic surgery following a positive test result; those with pessimistic personalities or a tendency towards emotional suppression; women younger than 40; and those familiar with severe consequences of breast and or ovarian cancer in their family. Lerman et al. (1994b) investigated anticipated responses to test results. A positive result was considered likely to induce anxiety, depression, impair quality of life but increase feelings of control. A negative test result was anticipated to lead to guilt and continued worry and desire for screening.

Case studies and anecdotal reports of the impact of genetic testing have shown the process to effect the whole family and to cause a number of complex emotional responses (Dudokdewit et al. 1997). In a study of 37 large high-risk families undergoing testing Lynch et al. (1999) reported variable and unpredictable emotional responses including sadness, anger, acceptance and relief from uncertainty in response to a positive results and relief, disbelief and 'survivor guilt' in response to a negative test result. Croyle et al. (1997) reported short-term effects during 1-2 weeks following BRCA1 testing. General distress was found to decline at follow up for all participants although carriers were found to show higher levels of general distress and test-related anxiety than non carriers. Lerman et al. (1996) conducted a 1 month

follow up of women undergoing predictive testing for breast cancer. Women with a negative result showed a reduction in depressive symptoms compared to those with a positive result. However women receiving a positive result did not show an increase in depression or functional impairment.

Broadstock et al. (2000) reported a lack of studies assessing the long term psychological consequences of predictive testing for late onset disease. There are concerns that genetic testing may actually reduce motivation for detection and prevention behaviour due to increased fatalism (Marteau and Lerman 2001, Senior et al. 1999, 2000). Lerman et al. (2000) reports an ongoing study looking at the effect of predictive BRCA1 and BRCA2 testing on mammography use at one year follow up. Few changes were observed in adherence to recommendations for women with either a positive or negative test result although longer follow up is required.

The majority of individuals who have undergone genetic testing to date have been in research families who have participated in genetic research for many years. Research families are generally Caucasian, well educated and have had much time to adjust emotionally to the new genetic knowledge before predictive testing could be offered. The research context of genetic testing has been conducted with regard for stringent ethical guidelines and incorporated protocols for extensive pre and post-test counselling (Broadstock et al. 2000). There are doubts about whether findings regarding the consequences of genetic testing can be generalised to newly identified women at increased risk who obtain genetic counselling in clinical practice where the processes of informed consent and genetic counselling are likely to vary from research protocols (Broadstock et al. 2000).

1.3.7 Prophylactic surgery

Women at high risk may consider bilateral prophylactic surgery as a preventative option. Interest in the procedure has been shown to be high in women at increased risk of breast cancer and the decision to undergo surgery has been positively associated with breast cancer worry, risk perception and biopsy history (Stefanek et al. 1995). Lerman at al (1996) reported that 17% of mutation carriers reported an intention to undergo prophylactic mastectomy. In a prospective study of 143 women

at increased risk who were offered prophylactic surgery Hatcher et al. (2001) found that the women who opted for surgery (n=79) had higher subjective perception of risk. Prior to surgery thirty-two percent of these women believed that they would inevitably develop breast cancer. Women who decided to undergo surgery were more likely to have had investigatory tests for previous breast symptoms or a genetic test than those who declined.

Retrospective studies of women who have undergone bilateral prophylactic mastectomy indicated that the procedure reduces emotional concern about developing breast cancer (Frost et al. 2000). A very small proportion of women have been shown to regret the operation and regret has been associated with lack of pre-operative counselling (Borgen et al. 1998). Prospective studies have indicated that women are satisfied with the outcome of surgery (Stefanek et al. 1995). Hatcher et al. (2001) found levels of general distress and anxiety were reduced at 6 and 18 months follow up for women who underwent prophylactic surgery but were sustained for women who declined surgery and were maintained on surveillance. Surgery was not found to have a detrimental effect on body image or sexual functioning. The majority of women in this study had immediate reconstructive surgery and the effect of different surgery on psychological response needs to be addressed in more detail.

1.4 DISTRESS IN WOMEN AT INCREASED RISK OF BREAST CANCER

A large proportion of women attending for genetic risk counselling are advised that they are at significantly increased risk of familial breast cancer but that their family history suggests genetic testing for BRCA1 or BRCA2 is not likely to be informative. There are concerns that ambiguous breast cancer risk information coupled with uncertain methods for detection and prevention may result in high levels of psychological morbidity in women with a family history of breast cancer. Women must face the fear of developing cancer along with its anticipated consequences of treatment and threat to survival as well as guilt about the possibility of passing a cancer predisposing gene on to their children. Distress may not only affect quality of life but also influence the cognitive processes involved in understanding risk information and decision-making about risk management.

A number of studies have therefore been carried out to determine and describe levels of distress in women at increased risk of breast cancer. Levels of distress in women with a family history of breast cancer have been reported to be higher than general population samples (Kash et al. 1992, Gagnon et al. 1996). The prevalence of psychological morbidity has been reported as comparable to that of breast cancer patients (Lerman and Schwarz 1993, Hopwood et al. 1998). However, other studies have reported that women with at least one first degree relative who have suffered from breast cancer show levels of distress comparable to controls without a family history of the disease (Wellisch et al. 1991, Lerman et al. 1994a, Lloyd et al. 1996, Zakowski et al. 1997).

Different levels of distress reported in these studies may reflect differences in sampling (ie extent of family history) as well as the timing and situation at which women were assessed. Levels of distress are likely to be higher for women attending clinics for risk counselling or screening. Women attending familial breast cancer clinics for the first time have shown levels of trait and state anxiety higher than controls but comparable to women attending for routine breast screening (Cull et al. 1999, Watson et al. 1999). Valdimarssdottir et al. (1995) assessed levels of distress in women attending a surveillance clinic the day before screening and one month later. Acute distress was higher prior to screening but reduced at follow up, whereas general distress was consistently higher than general population norms at both time points.

The prevalence of distress might vary depending on the assessment questionnaire used and cut off point chosen (Hopwood et al. 1998). It is worth remembering that screening questionnaires overestimate actual psychological morbidity (Coyne et al. 2000). Hopwood et al. (1998) compared the prevalence of psychological morbidity of 158 women assessed 3 months after genetic counselling determined by the GHQ and psychiatric interview. Twenty-six percent of women showed significant distress on the GHQ however the psychiatric interview confirmed psychiatric disorder in just 13% of women (Hopwood et al. 1998). This rate was still higher than would be expected in age-matched controls.

1.4.1 Types of distress

It has been suggested that global measures of distress fail to capture the specific sources of distress for women at increased risk of breast cancer and that measures of breast cancer related anxiety are more informative (Hopwood et al. 1998, Kent et al. 2000, Thewes et al. 2001). Models of response to stress have indicated that assessment of appraisals and emotions related to the specific situation are likely to be more informative than general measures (Lazarus and Folkman 1984). Different types of distress in this sample may require alternative interventions. Standard psychiatric interventions may be appropriate for women showing high levels of depression or anxiety whilst those with cancer related anxiety might benefit more from targeted psycho-education programmes (Thewes et al. 2001). Recent studies have therefore incorporated measures designed to assess anxiety specifically associated with breast cancer (Lerman et al. 1996, 1997, Audrain et al. 1997, Croyle et al. 1997).

A concept prevalent in the contemporary literature is cancer worry. This represents unwanted aversive thoughts and emotional discomfort specifically associated with cancer risk and is apparent for many women at risk of breast cancer (Kent et al. 2000). Worries about breast cancer have been frequently assessed in women at increased risk (Lerman et al. 1991, 1993, 1994a, Epstein et al. 1997) and women in the general population (McCaul et al. 1996, 1998). Studies have used Likert response items to address the frequency of breast cancer worries and the degree to which these concerns impinge on everyday life. From this literature a cancer worry scale has been developed (Watson et al. 1998) to assess worry directly associated with breast cancer risk. A cancer related anxiety and helplessness scale has also been utilised (Kash et al. 1992) although has not been published to date.

In a population study of women with at least one first degree relative with breast cancer Lerman et al. (1994a) found a third of women of all ages reported worries about breast cancer that interfered with their capacity to function in everyday life. Watson et al. (1999) found 28% of women attending a familial breast cancer clinic for the first time worried about breast cancer frequently or constantly and 18% felt this worry was a severe or definite problem. Hopwood et al. (2001) also reported that

two thirds of 500 women referred to genetic counselling for breast cancer reported high levels of worry about developing cancer in the future although few women reported these worries interfered with their mood or daily functioning. The authors argued that cancer worry might actually represent a realistic response rather than morbid anxiety (Hopwood et al. 2001).

Another measure of cancer specific distress widely used in the literature is the Impact of Event scale (Horowitz et al. 1979). This measure was originally developed in order to assess subjective levels of distress associated with a specific event in the context of the stress response syndrome. It measures two constructs: intrusion (thought, images dreams, waves of feeling) and avoidance (denial, blunted sensation, behavioural inhibition, emotional numbness). The measure can be tailored to assess distress associated with a specific event and has been adapted to assess intrusion and avoidance associated with breast cancer risk (Kash et al. 1992, Lerman et al. 1993, 1994a, Lloyd et al. 1996, Thewes et al. 2001). Zakowski et al. (1997) and Lloyd et al. (1996) reported that women at increased risk because of family history showed higher levels of intrusive thoughts about breast cancer and avoidance than controls without a family history. Lerman et al. (1994a) found over half of their sample reported intrusive thoughts and feelings about breast cancer risk and levels of intrusive thoughts were comparable to those observed in populations exposed to a traumatic stressor. Valdimarsdottir et al. (1995) also reported high levels of intrusive thoughts and avoidance of breast cancer in women on surveillance programme both assessed prior to screening and at the follow up 1 month later.

1.4.2 Impact of genetic counselling on levels of distress

A number of prospective longitudinal studies have been designed specifically to determine the impact of genetic risk counselling for breast cancer on levels of general and cancer specific distress in order to determine if genetic counselling reduces or invokes distress. Results have been inconsistent. Cull et al. (1999) reported that levels of general distress were reduced 4 weeks following genetic counselling for breast cancer and Julien-Deynier et al. (1999) showed a reduction in state anxiety following genetic counselling across studies in France. However, Hopwood et al. (1998) reported no change in prevalence of psychological morbidity

at 3 months following genetic counselling and Watson et al. (1999) reported no change in general distress at annual follow up.

A few studies have reported a reduction in cancer specific distress subsequent to genetic counselling. Gagnon et al. (1996) found a reduction in cancer worry at 4 months follow up and Hopwood et al. (2001) found a reduction in cancer worry following genetic counselling for a subset of women who initially overestimated their risk as their perception became more accurate. Other studies have reported no change in cancer specific distress including both cancer worry and intrusive thoughts at 1, 3, 6 and 12 months following genetic counselling (Kent et al. 2000, Watson et al. 1999). There are little data on longer term follow up of women although qualitative studies suggest that cancer specific anxiety is prominent in at least a proportion of women who have been attending a familial breast cancer clinic for over 2 years (Appleton et al. 2000).

1.4.3 Predictors of distress

It appears that there are large variations in levels of distress subsequent to genetic counselling and that subgroups of women maintain high levels of anxiety and worries about breast cancer. There is a need to determine why some women are more prone to high levels of anxiety. A clearer understanding of the cause of distress in this population will help to identify women requiring additional support and plan psychological support services.

Lerman and Schwarz (1993) suggest that possible determinants of distress might include situational factors (eg personal risk, disease related factors), personal factors (eg demographics), perception of the situation (eg risk perception) and coping efforts. Subjective perception of risk shows stronger and consistent association with levels of distress than does objective risk status (Lloyd et al. 1996, Kent et al. 2000, Zakowski et al. 1997). Women with higher risk perceptions or who overestimate their risk also show higher levels of cancer specific anxiety (Watson et al. 1999, Lloyd et al. 1996, Kent et al. 2000) and general distress (Audrain et al. 1997). Younger age has been associated with increased levels of general distress in women with a family history of breast cancer both in population based studies (Lerman et al.

1994a) and follow up assessments after genetic counselling (Cull et al. 1999). Personality factors have also been associated with levels of distress in this population. Audrain et al. (1997) found women who were less optimistic reported higher levels of general distress. Lerman et al. (1996) investigated the impact of coping style on response to genetic counselling and found that monitors who tend to seek out information in a threatening situation showed higher levels of distress after genetic counselling than blunters who attempt to avoid the situation. Audrain et al. (1997) reported that together monitoring and optimism accounted for approximately a third of variance in general distress in women who had self referred for genetic counselling for breast or ovarian cancer.

In the follow up assessment to genetic counselling Cull et al. (1999) also found that risk of distress was higher for women who had reported high levels of distress prior to attending the clinic. In psychiatric interviews of women referred for psychological help following attendance at a familial breast cancer clinic Hopwood et al. (1998) found factors contributing to levels of distress were often longstanding prior to attending for risk counselling. Problems were not necessarily associated directly with breast cancer risk and included work and financial problems, relationship problems and loss and unresolved grief (Hopwood et al. 1998). A number of authors have suggested that bereavement response and prior experiences of cancer in the family may contribute to levels of distress (Wellisch et al. 1992, Zakowski et al. 1997, Cull et al. 1999, Valdimarsdottir et al. 1995, Hopwood et al. 1998, 2001). To date there is limited information on the causes of distress in women at increased risk of breast cancer. Studies have tended to be descriptive in nature, identifying variables that are associated with or predictive of distress rather than testing causal explanations. Further research is needed to examine the cause of distress in a theoretical manner in order to test causal associations and identify factors that may lend themselves to intervention.

1.5 SUMMARY AND CONCLUSIONS

Breast cancer is a common disease in women although the aetiology is not fully understood. A number of factors have been identified as potential causes of the disease but few are amenable to personal control. The symptoms and progression of

breast cancer are variable and unpredictable. The timeframe of the disease is uncertain and may be protracted. Treatment may control localised disease but rarely cure advanced metastatic cancer and patients face a risk of spread and recurrence. Breast cancer is a distressing, stressful condition and the diagnosis and treatment of the disease may have a large impact on the patient and her family. Although detection methods and treatment strategies are improving, the efficacy of screening remains unclear in younger women and no fully effective means of prevention are available.

Increased understanding of the molecular genetics involved in breast cancer has lead to the discovery of heritable mutations that predispose carriers to developing breast cancer at an early age. Mutations in these genes account for a proportion of familial breast cancer cases although there are likely to be other genes yet to be discovered. Public awareness and concern has lead to the development of cancer genetic services aimed at providing risk assessment and health management to women with a family history of breast cancer. Variable methods are used to assess risk status and communicate risk information making the evaluation of the effectiveness of genetic counselling difficult. National guidelines for cancer risk management are currently under development.

Although interest in predictive testing for breast cancer is high only a small subset of women with a strong family history of breast cancer concordant with patterns associated with BRCA1 and BRCA2 mutations are eligible for predictive testing. Predictive testing is problematic because of the possibility of false negatives and uncertainty regarding the penetrance of the BRCA1 and BRCA2 genes outside research families. For the majority of women in the UK who are deemed at moderate or high increased risk of breast cancer genetic testing would not be informative. These women are provided with a risk estimate and offered regular screening in order improve early detection of the disease and reduce mortality. However the clinical efficacy and cost effectiveness of screening younger women with a family history of breast cancer remains controversial. The uptake of recommended screening practices is less than optimal and often associated with distress.

Women actively seek genetic counselling services because of concerns about breast cancer in their family. Women attending for genetic counselling must comprehend a great deal of complex medical information. However, there is much uncertainty concerning whether, when and where gene carriers will be affected. For women with a significant family history of the disease but for whom a specific mutation has not been identified the risk is even more uncertain. Based on this information women must make decisions about their health management and adjust emotionally to their risk status.

Concern regarding the impact of uncertain genetic risk information on mental health has prompted research into the psychosocial consequences of discovering and living with an increased risk of breast cancer. Levels of distress in women at increased risk are variable. It is unclear whether genetic counselling helps reduce levels of anxiety and cancer specific distress or has beneficial effects on mental health. A proportion of women attending familial breast cancer clinics have high levels of distress that are not alleviated by genetic counselling. A number of factors have been postulated to be associated with distress although few studies have examined these factors in detail.

It has been suggested that past experiences of breast cancer in the family contribute to psychological difficulties in women at increased risk of the disease. The subsequent two chapters will examine the literature surrounding prior experience of breast cancer in the family and psychosocial response to risk in more detail and outline a study to determine the effect of experience of breast cancer in the family on levels of distress in women at increased risk of breast cancer.

CHAPTER 2

EXPERIENCE OF BREAST CANCER IN THE FAMILY

EXPERIENCE OF BREAST CANCER IN THE FAMILY

2.1 INTRODUCTION

2.1.1 Psychosocial impact of cancer in the family

The occurrence of cancer has a large psychosocial impact on the family. Living with an affected relative can be even more difficult than coping with one's own illness (Hilton 1993). Emotional responses to a relative's diagnosis of cancer have been found to be similar to those of patients and include fear, anxiety, confusion and uncertainty (Leedham and Meyerowitz 1999). Cancer in the family can disrupt family routines and may force other family members to take on new responsibilities and family roles. This in turn can lead to resentment, tension and anxiety (Kelly et al. 1987). Cancer in the family also creates dilemmas regarding communication about cancer and the degree of involvement in the relative's medical care and personal issues.

Cancer in women can have a large impact on family functioning because women are often central figures in managing family life. Northouse (1995) reviewed the psychosocial impact of maternal breast cancer on the family. The mother's illness was found to have a significant effect on the emotional well being of the spouse and children, to disrupt family life and alter family roles (Northouse 1995). Distress in children has specifically been associated with the threat of loss of mother, temporary loss of mother during the illness, side effects of treatment and disruption in family roles and routines (Compas et al. 1996, Spira and Kenemore 2000).

Breast cancer is an extremely variable illness. The practical and emotional impact of the disease on the family is likely to depend on a number of illness factors including the type of breast cancer, how the disease was detected, treatment regime, disease progression and prognosis. Compas et al. (1996) found children's perception of the seriousness of parental cancer was associated with increased stress about the illness experience and greater emotional distress. Children were found to hold accurate appraisals of the seriousness of parental cancer compared with the actual stage and prognosis of the disease. In a qualitative study of the impact of maternal breast cancer on the family, Hilton (1996) found that visible and intrusive symptoms and/or

treatment made it difficult for families to return to normal. Veach and Nicholas (1998) suggested that the impact of cancer on the family depends on the phase of illness (prediagnosis (testing), diagnosis, treatment, rehabilitation and remission) although understanding of the psychosocial issues raised during these different phases is limited. Northouse (1995) found women suffering from recurrent breast cancer or those with high levels of symptom distress reported more problems with family roles and a stronger detrimental impact of the disease on the family.

A number of additional individual and family factors have been found to moderate the impact of the disease on the family. Veach and Nicholas (1998) suggested that the developmental stage of the family is an important factor and that different family types (ie new couples, young families, families with adolescents, ageing family) may be disrupted in different ways. Other factors found to be important include children's age, gender, the quality of the parent-child relationship, family coping behaviours and flexibility in role sharing, communication styles, financial resources and concurrent stressors (Compas et al. 1996, Hilton 1996, Lewis et al. 1993, Lichtman et al. 1984, Vess et al. 1985). Family members directly involved in caring for relatives with cancer may also experience increased levels of stress (Siegel et al. 1996).

Bereavement of a loved one is reported to be one of the most stressful life events a person will face and has been associated with a decline in both physical and mental health (Dohrenwend and Dohrenwend 1974, Kurtz et al. 1997). Bereavement from cancer has been reported to be particularly traumatic due to the prolonged and stressful nature of the illness (Koocher 1986). Loss of a primary family member from cancer has been associated with increased depression (McHorney and Mor 1988). The risk of depression following bereavement from cancer has been positively associated with a number of factors including younger age of patient, lower satisfaction with care giving and increased family tension (McHorney and Mor 1988).

Research to date has largely focused on the effects of cancer and bereavement on the spouse or young children of cancer patients. A wider view of the family needs to be

considered when investigating the impact of breast cancer when there is a familial predisposition. Research has also tended to focus on the impact of diagnosis and early stage breast cancer with limited work addressing the impact of other illness phases. The long-term effect of cancer in the family needs to be addressed utilising long-term prospective follow-ups. Retrospective studies have indicated that experiences of cancer in the family have long-term psychosocial implications and that unresolved issues may remain years after the illness experience (Kelly 1987, Leedham and Meyerowtiz 1999). Relationship difficulties in the family stemming from breast cancer have been shown to be long-lasting (Lichtman et al. 1984). Studies of relatives involved in care-giving have identified psychological distress and depressive symptoms following bereavement to be persistent (Siegel et al. 1996, Kurtz et al. 1997). Finally the experience of *multiple* illness and bereavement from cancer in the family has yet to be examined. Research on AIDS-related bereavement in gay men found that stress responses and depression increases with the number of bereavements experienced (Martin 1988, Gluhoski et al. 1997).

2.1.2 Psychosocial impact of familial cancer.

Women with a family history of breast cancer are likely to have a number of family members who have suffered from the disease at an early age (see Figure 1.1, page 29). Relatives are likely to have witnessed and been personally involved in the physical, emotional and social consequences of breast cancer including fear at the time of diagnosis, observations of the disabling and disfiguring consequences of treatment and emotional consequences of the disease in the family. These experiences may become even more threatening when women become aware of their own increased risk of developing the same disease. Women may therefore enter genetic counselling about their own risk with strong emotions and fears derived from cancer related experiences and losses within their family. McAllister (2002) reports findings from a grounded theory that proposes the process of 'engaging' with cancer risk (the degree of cognitive and emotional involvement with risk) is influenced by experiences of cancer within the family. Interviews with individuals at risk of Hereditary Non Polyposis Colorectal Cancer indicated that individuals who had witnessed suffering or death from cancer in their family were most intensively engaged with their risk (McAllister 2002).

A variety of illness, individual and family factors may create very different *subjective experiences* of breast cancer for women with the same *objective* family history. The subjective experience of breast cancer in the family is likely to have a large impact on how women think, feel and respond to their own risk of breast cancer and may help to explain the wide variation in response to risk information. The following section will review studies that have addressed the experience of breast cancer in women at increased risk. The review aims to describe the experiences of these women and highlight issues that may influence response to risk.

2.2 EXPERIENCE OF BREAST CANCER IN THE FAMILY: LITERATURE REVIEW

2.2.1 Experience of breast cancer: Qualitative studies

Anecdotal accounts have reported that witnessing breast cancer in close relatives has both negative and beneficial effects (Lynch et al. 1994). Emotional themes revealed include fear of death and identification with mutilated maternal body image, anger and bewilderment, unresolved grief and depression, guilt regarding lack of time spent with the relative and lowered self-esteem (Kelly 1983, Lynch et al. 1994, Wellisch et al. 1991, 1992, 1996). Experiences of breast cancer in relatives also inform women about the disease and motivate them to seek information and screening (Lynch et al. 1994).

2.2.1a Maternal breast cancer

Wellisch et al. (1991, 1992, 1996) conducted a series of qualitative studies examining psychological functioning in daughters of breast cancer patients. Participants were found to perceive a decline in their mothers' quality of life (self image, attractiveness and sexuality), social relationships and activities of daily living following breast cancer. Those who had lost their mother to breast cancer reported a greater deterioration of their mothers quality of life and daily activities. These perceptions were associated with poor psychological adjustment (Wellisch et al. 1996). Maternal breast cancer was also found to affect daughters self concept including self image and sexuality (Wellisch et al. 1991, 1992).

> *"Why should I get attached to my body and start enjoying sex when all that will be totally destroyed when I get breast cancer like my mother did?"* (Wellisch et al. 1991, pg 334)

Women reported that their experiences had altered their role in the family and long range life plans (Wellisch et al. 1992). Changes in life plans reflected changes in family life during the illness experience as well as changes in future plans for adult life (eg marriage plans). Some women with a family history of breast cancer plan their lives around the certainty that they themselves will develop the disease (Kelly et al. 1987). For example, in psychiatric interviews with women following genetic counselling for breast cancer Hopwood et al. (1998) reported two women who made the decision not to have children specifically to avoid their offspring having to face the loss of their mother as they had done.

Psychiatric interviews with women following genetic counselling have shown that loss of a mother due to breast cancer can result in unresolved grief that continues to affect some women for many years and may contribute to psychiatric morbidity (Hopwood et al. 1998). Anecdotal reports have indicated that learning about ones risk status can reactivate grief for lost relatives and may contribute to distress following genetic counselling for breast cancer (DudokdeWit et al. 1997, Lodder et al. 1999).

Wellisch et al. (1992) found that age at the time of the mother's diagnosis was also associated with psychological adjustment. Women who were adolescents (11-20 years) had more adjustment problems and showed poorer psychological well-being than women who were younger (0-10) or adults (20+). The adolescent group was also more likely to report discomfort about involvement with mother's illness, poor resolution of feelings about it and less subsequent satisfaction with their own sexual relationships. Clinical interviews with adolescent daughters of mothers with breast cancer have indicated anxiety about family roles and their relationship with their mother as well as fear of recurrence of mothers breast cancer (Spira and Kenemore 2000). This clinical sample showed fears concerning personal physical and sexual development because of their own breast cancer risk (Spira and Kenemore 2000). These issues may reflect difficulties confronting maternal breast cancer during a

vulnerable emotional and sexual developmental stage (Wellisch et al. 1992, Spira and Kenemore 2000).

2.2.1b Communication about breast cancer

Chalmers et al. (1996) reported that communication patterns within the family at the time of the relative's illness can influence adjustment and adaptation to personal risk. Communication styles that were restricted to the physiological process involved in breast cancer and which neglected personal and emotional aspects of the illness, combined with unrealistic optimism and positivity about the illness were fairly common. However, this communication style was associated with fear and anxiety about breast cancer, family conflict over the course of the illness, poor preparation for relative's death and bereavement problems.

2.2.1c Stages of adaptation

Chalmers and Thompson (1996) conducted interviews with 55 women with a first degree relative with breast cancer in order to understand how women's experiences impact on their response to their own cancer risk. A third had lost their relative to breast cancer, the remainder reported that their relative was alive and in remission. The study indicated that women need to come to terms with their relatives' illness in order to adjust fully to their own risk. Analysis revealed three stages of adaptation: living the breast cancer experience; developing a risk perception and adjusting to the personal risk of breast cancer. Women were described as having lived vicariously through their relative's breast cancer and to have shared the illness experience by developing an 'emotional connection' with the relative. The degree to which the experience was shared depended on the woman's age, relationship with the affected relative and concurrent stressors in the woman's life. Younger women were more likely to report that sharing the cancer experience resulted in greater fear and anxiety. Illness that was erratic, unpredictable and unresponsive to treatment was found to be more emotionally demanding than illness that responded well. Greater contact with the relative and more intense experiences (eg severe uncontrolled pain) appeared to increase vicarious experience. Intense breast cancer experiences were more difficult for the participant to resolve and hindered development of a personal risk perception. Risk perception was found to increase as the participant neared the age of their affected relative and was higher for women who felt they resembled their relative

both physically or in personality. Individual's feelings of susceptibility to cancer appears to increase as they near the age at which a relative was diagnosed or died from cancer and often prompt attendance at familial breast cancer clinics (Richards et al. 1995, Brain et al. 2000).

Chalmers and Thompson (1996) found some women able to integrate their personal risk perception with their sense of self and adapt to risk by developing a sense of personal control. This was achieved by self care practices such as screening, maintaining a healthy lifestyle and rehearsing how to cope with the development of cancer. This was not permanent state but altered with changing experiences surrounding breast cancer in the family. Not all women achieved this final phase.

2.2.1d Risk management decisions

The experience of family members can influence women's decisions about risk management, for example uptake of screening, considering genetic testing or prophylactic surgery. Dudok deWit et al. (1997) carried out a case study of a family in Holland who were involved in a genetic research study of Hereditary Breast and Ovarian Cancer (HBOC) and were the first family for whom predictive testing became available. The emotional response, family dynamics and individual's decisions about genetic testing and prophylactic surgery were investigated. Decisions to undergo predictive testing and prophylactic surgery appeared to be made following the death of relatives supporting the stages outlined by Chalmers et al. (1996).

> *"The worsened physical condition of her sister Mrs B. strengthened her pre-test opinion in favour of preventative surgery"* (Dudok deWit et al. 1997, pg 67).

Decisions about predictive testing or prophylactic surgery were also based upon the degree to which individuals identified with affected relatives particularly if they were of similar age at which a relative was diagnosed with breast or ovarian cancer.

"Three identified female gene carriers from the older generation chose to have a prophylactic ovariectomy, but no mastectomy. They viewed ovarian cancer as threatening because in their previous generation the women had died of ovarian cancer at the same age as they were now." (Dudokdewit et al. 1997, pg 67).

2.2.2 Experience of breast cancer and response to risk: Quantitative studies

The qualitative studies outlined above highlighted certain aspects of experience that may influence psychosocial response in women at increased risk of breast cancer. Quantitative studies are required to assess the impact of these experiences on women's response to breast cancer risk. Few studies have been designed directly to investigate the impact of experience on response to risk although some studies have included measures of experience that were not the main focus of the study. The limited work on this construct most probably reflects the difficulties in quantifying women's experience.

2.2.2a Exposure to cancer

Lodder et al. (1999) investigated psychological functioning in women waiting for test results for predictive testing for BRCA1 or BRCA2. Women who had more relatives affected with either breast or ovarian cancer and women whose relatives had suffered from cancer at a younger age (<40) reported higher levels of general and cancer specific distress. Baider et al. (1999) assessed women attending a one day conference on familial breast cancer. Although women were of unknown risk status and the sample was prone to selection bias the results indicated that women who reported that both a mother and sister to have been affected with the disease showed higher levels of intrusion than those with just one affected first degree relative.

Dudok deWit et al. (1997) assessed the effects of predictive testing for a range of late onset inherited disorders (including breast/ovarian cancer, Huntingtons disease, cerebral haemorrhage and bowel cancer). Individuals with greater experience of the disease in close relatives who reported that the disease had had a greater impact on their lives reported higher levels of distress prior to the test. Clear recollection of symptoms observed in affected relatives and emotional descriptions of the impact of the disease on their lives (including shame, fear, anger) were associated with higher levels of intrusive thoughts about the disease.

Lodder et al. (1999) also found that exposure to breast cancer in its later stages was associated with levels of depression. Women who knew/had known close relatives with metastatic cancer showed higher levels of distress compared to women who did not know of any such relatives. The authors suggest these experiences increase familiarity with the serious consequences of the disease and awareness of what the genetic test represents.

> *"I know what I am talking about, since I've seen cancer in close relatives too often. The sickness of these relatives and the cancer hospital come regularly to my mind and I don't want to end up there too"* (Lodder et al. 1999 pg 912)

Qualitative studies had indicated that maternal experience of cancer may be particularly distressing. Julian-Reynier et al. (1999) looked specifically at the impact their mothers' cancer on levels of distress in women prior and post cancer genetic risk assessment in cancer centres across France. Of 219 women assessed, 73% were undergoing genetic risk counselling because of a family history of breast cancer and 22% because of a family history of bowel cancer. Women whose mother had been affected with cancer showed significantly higher levels of anxiety one week post genetic counselling. Analysis of specific cancer types was not reported.

2.2.2b Bereavement from cancer

A few studies have assessed the impact of bereavement on women's psychological response to cancer risk. Wardle (1995) investigated the influence of the number of friends and relatives who had developed, or died from cancer, on risk perception of women at increased risk of ovarian cancer. Women at increased risk were compared with women who had taken part in community screening in the previous year and a group of controls who were not taking part in any ovarian screening programmes. Both screening groups had experienced more cancer deaths than the controls. In all three samples women who knew more people who had died of cancer reported a significantly higher personal perception of risk. A significant interaction indicated that the effect was significantly stronger in the screening groups than the controls. Bereavement from cancer has also been associated with depression. Thewes et al.

(2001) found that the total number of first and second degree relatives who had died from cancer was associated with levels of depression in women with a family history of breast cancer.

Zakowski et al. (1997) assessed the impact of parental bereavement on both women's perception of breast cancer risk and levels of distress. Two groups of women at increased risk of breast cancer, those who had, and had not, suffered a parental bereavement due to cancer were compared to a control group who had no history of cancer in their first-degree relatives. Levels of perceived risk differed between all three groups. Women at increased risk showed significantly higher risk perceptions than the control group. However, women who had lost a parent due to cancer had significantly higher perceived risk than women who had not.

Parental death from cancer also appeared to account for some of the variability in levels of distress. Women who had suffered parental bereavement due to cancer reported higher levels of cancer specific distress than those who had not. Women who had *not* lost a parent to cancer showed levels of distress comparable to the control group. This suggests that cancer related events influence psychological response to risk. The authors tested the hypothesis that perceived risk mediated the impact of experience of parental death from cancer on levels of distress. Multiple regression analysis confirmed this association. Women who had suffered a parental bereavement because of cancer showed higher personal perception of risk that appeared to account for the variability in levels of distress. The authors speculate that

> *"...the experience of the parent's death from cancer, in addition to their diagnosis which all women in our risk group had experienced, may change the meaning of cancer from a potentially curable illness to a death threat. This may heighten these women's distress about cancer in general" (Zakowski et al. 1997, pg 367).*

Although these studies provide initial evidence for the impact of experience of cancer on response to risk information neither study assessed the nature of these experiences in any detail. The measures used were basic and categorical assessing general cancer experiences such as the number of people known to have cancer, or whether the women had suffered a parental bereavement due to cancer or not

(Wardle 1995, Zakowski et al. 1997). Wardle (1995) did not focus specifically on experiences in the family and included experiences of cancer in friends in the analysis. The sample size in the study by Zakowski et al. (1997) was too low to assess the effects of parental bereavement specifically from breast cancer. Previous research had suggested that it is the specific experiences of *breast cancer in the family* that influence women's response risk (Wellisch et al. 1991, 1992, 1996, Chalmers and Thompson 1996).

Hopwood et al. (2001) investigated the impact of maternal bereavement from breast cancer on levels of distress. In a study of 330 women assessed prior to genetic risk counselling for breast cancer no difference was found between women who had lost their mother to breast cancer (33%) and those who had not. In contrast to this finding, Erblich et al. (2000) found women with a family history of breast cancer who had suffered a maternal bereavement showed higher breast cancer related distress in terms of intrusive thoughts and avoidance than those who had not. Maternal death was associated with higher distress even though the average time since death in the sample was 14 years. Women who had been involved in caring for their ill mother also showed higher levels of cancer specific distress than those who had not. This may reflect stress of caregiving in general or increased exposure to the disease and its consequences. Alternative types of caregiving (eg physical help/emotional support) may have different effects. Women who had experienced both caregiving and maternal death showed the highest levels of distress and depressive symptoms. In contrast to the findings of Zakowski et al. (1997) the effect of these experiences on levels of distress was not found to be mediated by perceived risk.

2.2.2c Age at maternal diagnosis/death

Zakowski et al. (1997) found no effect of age at parental death, recency of bereavement, or parent's age at death on levels of distress although this analysis was limited by a small sample size (n= 30). Erblich et al. (2000) found no effect of age at maternal diagnosis or recency of diagnosis on of levels of distress in women with a family history of breast cancer. Hopwood et al. (2001) extended the work by Wellisch et al. (1992) on maternal bereavement during adolescence. Women who

had lost their mother between the ages of 10-20 (n=43) showed slightly higher levels of distress prior to genetic counselling than those who were bereaved after the age of 20 (n=115), or not at all, although this difference was not significant. Surprisingly women bereaved at the youngest age (<10) (n=24) showed significantly lower levels of cancer worry than the other age groups and were less likely to overestimate their risk.. It is possible that children of this age may have been protected from the impact of cancer in the family. The low number of participants in this sub-sample hinders the reliability of the result.

2.2.2d Experience of other unaffected family members

As well as the experience of affected relatives the experiences and decisions of other healthy family members may influence individuals' response to their own risk. For example Dudokde Wit et al. (1997) found that the first utiliser of options such as genetic testing and preventative surgery became the example for the rest of the family. Smith et al. (1999) assessed the impact of siblings' test results on psychological response to predictive testing for BRCA1. Test related distress was assessed 1-2 weeks following test results. Women who tested positive showed higher levels of distress at follow up than those with negative test results. However, psychological reaction to personal test results was found to be moderated by siblings' results. Those whose siblings all tested negative or whose sibling had not been tested experienced the greatest level of distress, exceeding scores of cancer patients 10 weeks after diagnosis (Smith et al. 1999). Men who were the first to be tested or non-carrier men whose siblings all tested positive also showed significant levels of test related distress compared to other men. These results suggest that family context is an important determinant of response to predictive testing. The authors also propose that a number of other factors may also be important, including how siblings respond to their test result, closeness of the relationship between siblings and the carrier status of other relatives.

2.3 SUMMARY

Cancer has a large psychosocial impact on all family members. The illness is distressing in nature, leads to changes in family functioning and roles and encompasses the threat of bereavement. Women at increased risk of breast cancer are

likely to have witnessed a number of relatives suffer from the disease at a relatively young age. Experiences of breast cancer in the family can vary widely given the diverse and unpredictable nature of the disease as well as individual and family differences in response to illness. The subjective experiences of two women with the same objective family history may therefore be very different. A family history of breast cancer may be particularly traumatic because of the associated personal risk of developing the disease and passing on an inherited susceptibility to one's children. Experiences of breast cancer among other family members are likely to have a strong influence on personal response and adjustment to risk status. However, studies on psychosocial response to risk have tended to focus on the individual in isolation from their family (Bottorff et al. 1998).

Qualitative studies have identified aspects of breast cancer experience that may influence psychosocial adjustment in women at increased risk. These include the age at time of relative's illness, pathogenesis of the disease, maternal bereavement, perception of ill relatives, vicarious living of the breast cancer experience, changes in life plans and family roles created by breast cancer in the family, identification with the relative and communication about breast cancer within the family.

Few quantitative studies have attempted to clarify the effects of experience on psychosocial adjustment in women with a family history of cancer. Greater exposure to the disease, in terms more affected relatives and closer experience of late stage illness, has been associated with increased levels of cancer specific distress and personal perception of risk. A few studies have also shown that experience of parental bereavement including maternal bereavement from breast cancer is associated with heightened breast cancer specific distress. Studies examining the effect of age at parental diagnosis or bereavement on subsequent response to risk are inconsistent. These studies prove difficult to compare because of the range of experiences often assessed within the same study (i.e. both maternal and paternal cancer experiences) and the lack of statistical power to examine specific effects. The impact of age at diagnosis and bereavement on response to risk is also likely to be moderated by a number of additional factors including family stage, involvement in care-giving and closeness to relative. The specific effects of these factors need to be

examined systematically to explore inconsistencies in the literature. Comparisons between studies are also hindered by assessment of diverse aspect of cancer specific distress. For example, certain experiences may have different effects on intrusive thoughts about breast cancer than on levels of cancer worry.

A number of other aspects of experience that may be associated with cancer specific distress have been highlighted in these studies including, recollection of symptoms, experiences of the consequences of the illness and care giving. Women may also be influenced not only by affected relatives but also the risk status and response of other healthy family members. These experiences may not only affect emotional response to risk but have also been found to stimulate information seeking and to influence decisions about management options.

The studies to date have been limited to basic categorical measures of experience (ie the experience of bereavement or not, caregiving or not). No quantitative study to date has investigated the subjective nature of these experiences such as women's interpretation and perceptions of their experience. This may be crucial in understanding the effect of experience on psychological adaptation to risk. Further quantitative studies are required to explore subjective experiences of women with a family history of breast cancer. Some pilot work to refine a locally developed questionnaire for use in this PhD project is described in Chapter 6.

The studies reviewed have been descriptive in nature aiming to identify salient aspects of experience without theoretical reflection. There has been limited consideration of mechanisms by which experience may determine psychosocial response to risk. There is a growing need to utilise a theoretical approach in order to clarify *how* experiences of breast cancer might influence risk perception, behaviour and the emotional well being of these women. This might then provide implications for change and intervention. An overview of theoretical perspectives on these issues is outlined in Chapter 3.

CHAPTER 3

THEORETICAL PERSPECTIVES

THEORETICAL PERSPECTIVES[2]

3.1 INTRODUCTION

The literature reviewed in Chapter 2 indicated that subjective experience of breast cancer in the family may be an important determinant of psychological well-being in women with a family history of the disease and may help explain variation in response to genetic risk information. The majority of research investigating the impact of women's experience of breast cancer has been exploratory and descriptive with limited theoretically driven work aimed at predicting psychosocial response to risk. However there are strong theoretical reasons for believing that aspects of experience of cancer in the family may be important predictors (Rees et al. 2001). Research driven from a theoretical perspective is able to identify and test hypotheses regarding potential causal mechanisms. Understanding of these processes could assist clinicians in predicting which women may exhibit high levels of distress or hold misperceptions of risk and increase understanding of factors influencing women's decisions regarding risk management. This holds important implications for service development and interventions designed to improve women's adjustment to risk. This chapter therefore aims to outline *how* women's experiences of breast cancer in their family may affect psychosocial response to risk from a number of theoretical perspectives.

The first section will outline theoretical perspectives on bias in risk perception and discuss potential cognitive bias in the interpretation of genetic breast cancer risk information that may be invoked by women's experience of illness in the family. The second section will describe models of decision-making and highlight how experiences of cancer in the family may influence decisions women make about managing their personal risk. The third section will outline how representations of health threats may influence psychosocial response to risk information. The impact of women's experiences on representations of breast cancer and subsequent emotional and behavioural response to risk will be discussed.

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[2]Material from this Chapter has been published:

Rees, G. Fry, A., and Cull, A. (2001) A family history of breast cancer: women's experiences from a theoretical perspective. *Social Science and Medicine* (52), 1433-1440.

3.2 COGNITIVE BIAS IN RISK PERCEPTION

In the general population individuals have been shown to demonstrate 'unrealistic optimism' when estimating their vulnerability to health threats. Individuals systematically underestimate their own risk compared to their perception of the risk of others (Weinstein 1980, 1982, 1987). Unrealistic optimism has been shown in women's perceptions of breast cancer risk (Absetz et al. 2000). Absetz et al. (2000) found Finnish women who were participating in, or waiting to join a screening programme for breast cancer perceived their own risk of breast cancer as lower than that of their peers. Research has indicated however that greater experience of the health threat in question (either directly or within one's social environment) and perceived similarity to disease victims reduces optimistic bias (Weinstein 1989, Lek and Bishop 1995). Indeed, Absetz et al. (2000) found optimistic bias was reduced in women with a first degree relative (FDR) who had been affected with breast cancer. This sub-sample showed no significant difference between perceptions of their own and peers risk of breast cancer. Absetz et al. (2000) suggests that the *experience* of having a FDR with breast cancer had a greater impact on risk perception than knowledge of this as a risk factor for the disease. Tversky and Kahneman (1974) describe three heuristics that create bias in risk perception when reasoning with uncertain probabilistic information. These heuristics have been demonstrated in genetic counselling situations (Shiloh 1994, Shiloh and Saxe 1989) and may explain how experience of breast cancer heightens personal risk perception.

3.2.1a Availability:

Easily recalled events (those which are more salient, familiar, recent and imaginable) are judged as more probable. Experiences of breast cancer in family, friends, work colleagues and the media have been associated with increased perceptions of risk in the general population (van der Plight 1998, Helzlsouer et al. 1994). For women at increased risk, frequent contact with affected relatives or recent experiences of breast cancer in their family will lead women to think about the disease more often and bring images of breast cancer to mind with greater frequency and clarity, heightening perception of personal risk.

3.2.1b Anchoring and adjustment:

Individuals are biased towards a preconceived idea about risk ('anchor') when provided with new risk information. Women may hold preconceptions of personal risk based on their experience that act as an 'anchor' from which new risk information given at the clinic is interpreted. For example a minority of women with a family history of breast cancer believe they will inevitably develop the disease prior to genetic counselling and this belief is maintained even after the genetic counselling session (Cull et al. 1999). The proportion of family members affected by breast cancer may contribute to women's expectation of their level of risk:

> *"I think it's inevitable because there's no female members of my family who haven't had it.....It's scary". (Appleton, 1999 quote from telephone focus group study of women at increased risk of breast cancer, personal communication).*

3.2.1c Representativeness:

Information about similarity and stereotypes are used to make judgements. Emphasis is placed on perceived similarities when judging probabilities. Women who feel they resemble an affected relative in their family may feel more vulnerable to the disease.

> *"I felt that I would get breast cancer as my body was similar to my mother's in many ways- she had fibroids and a hysterectomy- she had gall stones and had her gall bladder removed. I had both these operations by my late thirties".* (J.Zatz 1996, Daily life and the new genetics: some personal stories, pg 28)

3.2.2 Genetic risk and bias

Understanding of familial disease risk is further complicated by misconceptions about genetics. Lay concepts of inheritance are often based on resemblance and beliefs that multiple characteristics are inherited together (Richards 1996). There is a tendency to believe that susceptibility to illness is inherited along with other personality and physical dimensions (Richards and Ponder 1996). These beliefs often persist even after scientific, Mendelian accounts of inheritance have been provided (Richards and Ponder 1996). Beliefs about the inheritance pattern in the family and resemblance to affected relatives may have a strong impact on perception of risk.

> *"When I was young my mother attributed her own breast cancer diagnosis to birth order. She talked about being the affected first born daughter of an affected first born daughter of an affected first born daughter. She told me that as a first born daughter in this line, I should expect to encounter the disease as well. With the diagnosis of one of my mother's younger sisters when I was 25, my mother stopped talking about the disease as a problem for first born daughters. Instead she dwelt on the personality traits that her affected sister shared with their mother- a certain intensity and vulnerability to stress looming large among them".* (Ellen Macke 1996. Daily life and the new genetics: some personal stories, personal stories pg 32).

It is also widely believed that a greater proportion of inheritance is acquired from the same sex parent (Richards and Ponder 1996). Women with a paternal family history of breast cancer are under-represented at genetic clinics (McAllister et al. 1998). It has been suggested that women with a paternal family history have lower perceptions of risk because of limited understanding of how a predominately female disorder can be passed on by males (Green et al. 1997).

3.3 DECISION MAKING AND BEHAVIOUR

Studies have indicated that experiences of breast cancer in the family and the responses of other family members to their risk may influence women's decisions about risk management (see sections 2.2.1d and 2.2.2d pages 63 & 68). Early studies of human reasoning and decision-making attempted to discern processes that individuals use in order to reach optimal decisions. Based on the 'utility theory' individuals were thought to make decisions by assessing the probability and utility (importance) of events in order to maximise positive outcome (von Neumann and Morgenstern 1947). Evidence for this rational decision-making strategy has remained controversial (Neumann and Polister 1992). It is now accepted that such decisions are based upon a *subjective interpretation* of both probabilities and utilities. The theory was therefore revised as the subjective expected utility theory (SEU) which proposes that optimal decisions are based upon the decision maker's personal expected utility of various outcomes.

Wroe et al. (1998) assessed subjective reasons for undergoing predictive testing in hypothetical scenarios and in individuals who had contemplated genetic testing for a

variety of disorders. In support of the SEU theory the decision made was predicted by the ratio of relevant personal reasons for and against testing. Emotions were considered as pros and cons when contemplating genetic testing. Both initial emotional reactions to genetic testing and emotions anticipated after testing appeared important (Wroe et al. 1998). Lerman et al. (1995b) found the main reason against genetic testing for breast cancer was concern about emotional reactions. Women anticipated anxiety, depression and impaired quality of life following a positive test result. Women's experiences of breast cancer in their family and their beliefs about the disease are likely to have a large impact on how they anticipate their response to a positive test result.

Theories aimed at understanding health-related behaviour have been developed from the general decision-making perspective. A number of models have been developed based on the premise that individuals make a rational analysis of the costs and benefits of possible behaviours. The best known model of health-related behaviour is the Health Belief Model (HBM). This was initially designed to explain and predict compliance with preventative behaviours such as screening and immunisation (Rosenstock 1966, Becker 1974). According to this model readiness to engage in health behaviour depends on four beliefs: perceived susceptibility to the health threat; perceived seriousness of the health threat; perceived benefits of action; perceived costs of the behaviour. An individual will be most likely to engage in preventative behaviour if they regard themselves as susceptible to a serious illness and consider some preventive behaviour to have more benefits than costs. Once an individual is ready to act, behaviour is triggered by cues (Rosenstock 1966). Cues may be internal, such as bodily states, or extraneous (eg, health messages in the media).

Experiences of breast cancer in the family are likely to influence several components of this model. Section 3.2 discussed the potential impact family experiences of breast cancer on perceived susceptibility to the disease. Experience of breast cancer in the family has also been found to alter perception of its severity and consequences (Wellisch et al. 1996). Beliefs concerning the efficacy of screening (BSE, CBE and mammography) may also be influenced by the impact of these techniques on detection of breast cancer in affected relatives. Finally, exposure to cues concerning

breast cancer risk may vary depending on cancer related events and communication styles in the family. For example, Benedict et al. (1997) found frequency of breast self examination was positively associated with how much daughters talked to their mother about breast cancer.

Components of the HBM, including perceived susceptibility and barriers to action, have predicted breast self examination among women in the general population (Champion 1987) although HBM has been used with limited success to explain poor adherence to screening in women at increased risk of breast cancer (eg Kash et al. 1992).

A more general theory of behavioural decisions that has been applied to health related behaviours is the Theory of Reasoned Action (TRA) (Ajzen and Fishbein 1980). This model suggests that intentions to perform a particular behaviour arise from both personal attitudes towards the behaviour and social influence. Attitudes towards the behaviour may be positive or negative depending on the perceived consequences of the behaviour. Social influence, known as 'subjective norm' refers to the perceived expectations of important others. The theory was later extended to the Theory of Planned Behaviour (TPB) with the addition of 'perceived behavioural control' in order to help explain behaviour that is not entirely under volitional control (Ajzen 1991). The model proposes that intention to perform behaviour will be strong if an individual holds a positive attitude towards the behaviour, perceives him/herself to have control over the behaviour and believes that significant others expect him/her to perform it.

Whilst this model has not been explicitly tested in women at risk of breast cancer, studies suggest that relatives may actively encourage or dismiss utilisation of genetic services. For example, women have been found to attend familial breast cancer clinics following advice from another family member (Brain et al. 2000) and to consider genetic testing following the experience of other family members (Dudokde Wit et al. 1997). The beliefs of family members and communication styles within the family may influence both attitudes towards the behaviour as well as providing strong subjective norms.

Miller et al. (1999) discuss the implications of utilising social cognition models to understand women's decisions regarding prophylactic oophorectomy. Miller et al. (1999) suggest that the models could be used as a framework from which to systematically explore the psychological consequences of all options available. They propose that tailoring decision-making in this way will enhance informed decision-making as well as satisfaction, quality of life and adherence in the longer term.

Although these models highlight the role of cognitive factors in response to risk, application of these models has been limited by poor definition of constructs and inadequate assessment measures (Strecher and Rosenstock 1996). Constructs are often neglected because of methodological difficulties in measurement (for example the cues to action from the HBM) and studies often focus on the comparative impact of constructs rather than testing the integrated model as a whole (Harrison et al. 1992). Little work has addressed interactions between constructs. The models also provide limited insight into the development of cognitions. The HBM specifically does not explain *how* perceptions are formed or translate into behaviour change and none of the models explain how perceptions are evaluated and modified. This makes the models static in nature and unable to account for dynamic changes over time.

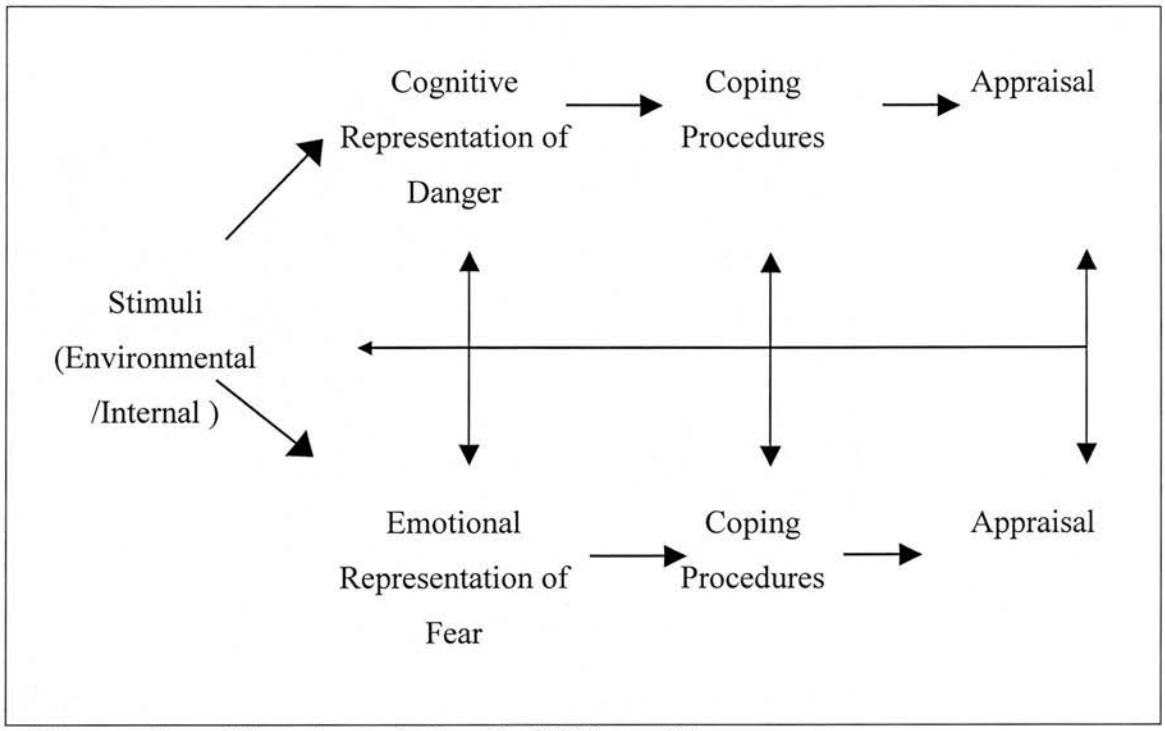
The models do not explicitly address emotional aspects of health threats and are therefore unable to account for biases such as denial and avoidance. Although the models were initially designed to inform understanding of preventative behaviours their application to detection behaviour (e.g. BSE) is limited without incorporating emotional factors. Although these behaviours convey benefit of early detection and treatment they are also anxiety provoking due to fear of discovering cancer. *Reasoned* perceptions regarding cancer detection can not be dissociated from emotional response. A wider dynamic model that is able to incorporate meaning and emotional response to risk is required to increase our understanding of psychosocial response to genetic risk of breast cancer.

3.4 ILLNESS REPRESENTATIONS

3.4.1 The Self Regulatory Model

Leventhal's (1980) Self Regulatory Model (SRM) takes into account individual's emotional and cognitive response to a health threat and aims to describe and predict how individuals respond to and cope with threats to health. The model was developed from work investigating the impact of fear on attitude and behaviour change, which indicated that cognitive and emotional aspects of health threats are processed independently (Leventhal et al. 1997). The model proposes that individuals actively generate cognitive and emotional representations of health threats and these representations independently guide and regulate behaviour. Illness representations are also known as illness perceptions and these terms will be used interchangeably throughout. The SRM is shown in Figure 3.1. As the model indicates stimuli from both the environment (eg risk information) and internal stimuli (eg the experience of symptoms) trigger cognitive and emotional representations. These representations may have been generated from past personal experience including actual experience of illness or vicariously from the experiences of friends and family, as well as from cultural beliefs and ideas inherent within language. Individuals derive an action plan to cope with threat based on their representation of it. The success of a particular coping strategy is appraised and feeds back into both the representation and the action plan, which may be modified accordingly. To give an example, an individual suffering from a severe headache may attribute this to stress and take medication for the pain. However, if the pain is not reduced and the headache worsens the individual may consider that the headache is a symptom of a more serious condition. The individual may become anxious and make plans to see their doctor. In this way both the representations of the health threat and coping plan have been modified following feedback from outcomes of the initial plan.

Figure 3.1- The Self Regulatory Model (SRM).



(Figure adapted from Leventhal et al. (1997), pg 21)

The SRM expands the social cognition models by integrating cognitive, emotional and behavioural elements and can be applied to a range of psychosocial outcomes (Leventhal and Cameron 1987). The model provides clear mechanisms by which experiences and conceptual knowledge may impact on beliefs and emotions in a dynamic manner. The model also allows for the influence of individual differences in how people represent, cope and appraise their response to health threats (eg coping style, attributional style). The SRM therefore provides a relevant framework from which to understand women's response to breast cancer risk. Women with different experiences of breast cancer will construct different representations of the disease and choose different coping strategies for dealing with risk and criteria from which to appraise their situation.

3.4.2 The nature of illness representations -qualitative assessment

Initial evidence for the SRM was gained from open-ended interviews with patients with hypertension and cancer (Leventhal et al. 1980, 1986, Meyer et al. 1985). These studies indicated that patients actively build a mental representation of their illness and that the illness representations were based around 4 components. These were: identity of the threat (its symptoms and label); cause (e.g. infection, genetic, stress); time line (duration and development) and consequences (including somatic and psychosocial). Lau and Hartman (1983) also found college students provided information concerning the control/cure of illness when asked about their most recent illness episode. Together these five components appear to capture the majority of comments made when people are asked to describe illness (Bishop et al. 1987).

The dimensions outlined in the SRM are not independent but likely to be highly interrelated. The dimensions are also not exhaustive and each attribute may be further differentiated (Nerenz and Leventhal 1983). An individual's model of an illness may also not necessarily be complete, well organised or medically correct. In this flexible manner the 5 dimensions outlined in the SRM provide a useful framework from which to investigate how individuals representations of disease are organised, structured and impact on response to illness.

Direct experience of illness has been highlighted as a main determinant of illness representations in affected patients (Schiaffino and Cea 1995, Paterson et al. 1999). To date there has been little research examining factors associated with the development of illness representations in healthy individuals. However Bishop and Converse (1986) and Bishop et al. (1987) provide evidence that healthy individuals hold representations of disease based around 'prototypes', i.e. 'idealised conceptions' that help organise information about illness and assist healthy individuals to match and evaluate symptom experiences. Bishop et al. (1987) found that 90% of healthy individuals' comments regarding illness fell into the 5 categories previously reported in patients. The most prominent dimensions healthy individuals spontaneously expressed were symptoms, labels and causes of illness.

Initial work on illness representations used open ended data collection in order to allow patients to describe and define their own representations. In a review of studies assessing illness perceptions between 1985-1995 Scharloo and Kaptein (1997) found that most researchers continued to use open ended or semi-structured interviews to elicit illness perceptions. Researchers most frequently developed codes or categories from recorded interview material although a number of studies combined interviews with predetermined study specific rating scales.

Qualitative methodology has the advantage of tapping representations directly without forcing participants to respond to predefined categories. However the disadvantages include the time consuming nature of such methods leading to low sample sizes and difficulty generalising results. Social desirability bias may also confound interview data. In a discussion of assessment of illness representations Leventhal and Nerenz (1985) note that even open ended interviews may not necessarily capture a representative illness perception but rather the most accessible responses at that moment in time. The authors also warn how questions may be subtly biased. For example the question '*How can you tell your blood pressure is high?*' is likely to elicit responses about *symptom* responses whereas the question '*How could your wife tell if your blood pressure was elevated?*' is more likely to evoke beliefs about the *behavioural* signs of hypertension (Leventhal and Nerenz 1985, pg 539). It is also difficult to determine the prevalence of beliefs and to investigate the relations between dimensions with qualitative data. The variability in qualitative methodological procedures and use of study specific rating scales also make comparisons between studies difficult.

3.4.3 Quantitative measurement of illness representations

Quantitative measures have the advantages of allowing researchers to explore the prevalence of beliefs, test the predictive value of combinations of illness dimensions, and to clarify issues regarding the conceptual overlap of dimensions. However Scharloo and Kaptein (1997) found studies utilising quantitative measures tended only to assess a limited number of dimensions. The most frequent dimension assessed was beliefs about control, measured in over 50% of the studies reviewed. A total of 22 different instruments were used to assess control beliefs across these

studies making comparison difficult. Study specific measures are also often used with limited validation work. Standard measures of illness perceptions are required in order to allow researchers to assess all dimensions in the same format, to compare perceptions of different illnesses, to investigate interrelations between dimensions and to assess the predictive value of patterns of illness representations as well as individual dimensions.

The Implicit Models of Illness Questionnaire (IMIQ) is a 38-item questionnaire designed to assess the dimensions of illness perceptions across a broad range of illnesses (Turk et al. 1986). It was developed to determine if there was a generic structure of illness representations across different illnesses (Turk et al. 1986). Turk et al. (1986) used the IMIQ to assess perceptions of diabetes, flu and cancer held by students, nurses and patients. The factor structure that emerged from these data suggested that a different structure underlies cognitive representations than had been previously proposed. The results suggested that illness representations are based on 4 dimensions: seriousness (permanent-chronic); personal responsibility; controllability and changeability (in terms of pain and disability). Schiaffino and Cea (1995) also used the IMIQ to investigate perceptions of rheumatoid arthritis, multiple sclerosis and HIV held by patients and students but reported a different factor structure. The results of these studies therefore suggested that the items did not to hang together in a consistent theoretically predicted manner. The application of the questionnaire across different illness is also questionable given the highly specific nature of some items (eg '*similar to the common cold*'). Factor analytic studies are only ever as good as the items entered into the analysis and the structures obtained are likely to change drastically depending on items used and the applicability of the items to the disease in question. Turk et al. (1986) themselves caution that '*the factors that we extracted had more to do with the questions that we asked than any cognitive representation of disease held by subjects*' (pg 471).

Heijmans and de Ridder (1998) also used factor analysis to investigate the structure of illness representations held by patients suffering from two different illnesses: Chronic Fatigue Syndrome (CFS) and Addisons disease. A different factor solution was found for each illness. For CFS, the factors appeared to represent manageability

(identity and control), seriousness, personal responsibility and external cause. For Addisons disease, the factors were interpreted as seriousness, cause, chronicity and controllability. Although the factor structures identified in these studies do not exactly fit the structure of illness representations described in the initial qualitative studies the types of dimensions described are similar and "*captured to some extent the spirit of the common sense model*" (Schiaffino and Cea 1995).

It is entirely consistent with the SRM that the structure of illness representations and importance placed on the dimensions differs depending on the disease in question (Schiaffino and Cea 1995). The factor analytic studies therefore do not dispute the 5 dimensions outlined by Leventhal, but indicate how illness perceptions may be grouped or organised for different illnesses. Although it may be difficult to compare alternative approaches (ie qualitative and quantitative) to investigating the structure of illness representations, authors have noted that content analysis of qualitative illness descriptions and factor analysis of quantitative data are *complementary* methodological approaches that are *likely* to elicit different results in this area (Lau et al. 1989, Bishop 1991). These approaches can be used to investigate different questions regarding the structure of illness representations. Content analysis of dimensions focuses on common features of illness representations whereas factor analytic techniques assess differences in the conceptualisation of diseases (Lau et al. 1989).

3.4.4 The Illness Perception Questionnaire

A more recent quantitative instrument designed to assess illness representations is the Illness Perception Questionnaire (IPQ) (Weinman et al. 1996). The authors aimed to develop a theoretically based, psychometrically sound assessment of illness perceptions that could be adapted to specific illnesses. It was hoped that such a measure could increase understanding of illness related coping and aid the development of interventions to improve self-management in chronic illness (Weinman et al. 1996). Items were derived theoretically to assess each of the 5 components of illness representations (identity, timeline, consequences, cure/control, and cause). The identity scale comprised a 12 item symptom checklist which can be expanded for specific illnesses. The timeline, consequences and cure/control scales

are assessed by items rated on a 5 point scale from strongly disagree to strongly agree. Participants also rate their beliefs about specific causes of the 5-point scales.

In order to check that the questionnaire adequately covered participants' representations, responses to the IPQ were compared with semi-structured interviews for a sample of 52 insulin-dependent diabetic patients. Participants completed both the IPQ and interview in a counterbalanced manner. All themes that emerged during the interviews were also apparent in the questionnaire data. In fact the IPQ was found to be more inclusive than the interviews since a third of respondents failed to provide information on some of the dimensions within the interviews. This confirms the findings of Leventhal and Nerenz (1985) who reported that checklists of symptoms and signs elicited more responses from patients with hypertension or cancer. This may reflect the fact that questionnaires are less likely to be influenced by situational and personality factors (ie articulation) than interview methodology. In a systematic review of causal attributions for heart disease French et al. (2001) compared patterns of responses elicited from open ended interviews or questionnaire methods. No differences were found in the frequency or rating of causal items when these items were generated by the respondent or the experimenter.

Weinman et al. (1996) conducted a thorough test of the psychometric properties of the IPQ on a number of patient populations including diabetes, rheumatoid arthritis (RA), renal, asthma, chronic fatigue syndrome (CFS) and myocardial infarction patients (MI). Internal consistency and test re-test reliability scores for each of the scales (identity, timeline acute, consequences, control/cure) were high. Intercorrelations between the subscales were also logical. Participants with a stronger illness identity were more likely to believe that their illness was long-lasting with greater consequences. Participants with less belief in the controllability of their illness were more likely to believe the illness was long-lasting with greater consequences. The IPQ showed adequate discriminant validity. The identity, consequences and timeline subscales as well as a number of cause items significantly differentiated between different illnesses (chronic pain, RA and CFS). Concurrent and predictive validity was tested on the MI sample. Subscales showed logical correlations with measures of perceived health and disability at baseline as well as

follow up measures at 3 and 6 months including recent doctor visits, self rated health, beliefs about likelihood of future MI and self rated beliefs about control over health problems. Since publication the IPQ has been used in a range of studies assessing patients response to heart disease, RA, psoriasis, chronic obstructive pulmonary disease, CFS and Addisons disease (Moss-Morris et al. 2002). The IPQ has also been adapted to assess the perceptions of healthy spouses of patients to enable investigation of the concordance between spouses' and patients' illness perceptions (Weinman et al. 1996, Heijmans 1999).

Original work on the IPQ showed that one of the subscales (cure/control) had lower internal reliability and closer investigation revealed that there were two separate components to this scale: personal control and perceived efficacy of treatment (Weinman, personal communication). In the review of assessment of illness perceptions Scharloo and Kaptein (1997) noted that the chronic pain literature suggested that the timeline subscale may be further differentiated since perceived constancy of the illness was considered an more important predictor of outcome than the perceived duration of illness in this sample. Moss-Morris et al. (2002) discuss potential improvements to the instrument and provide a revised version of the questionnaire (IPQ-R). The control/cure subscale was subdivided to create separate subscales to assess beliefs about personal control and perceived efficacy of treatment ('treatment control'). The timeline subscale was expanded and a measure of beliefs about the constancy of illness was developed ('timeline cyclical' subscale) along side the subscale to assess duration of illness (renamed 'timeline chronic/acute'). In addition, two further components of the theoretical model were introduced. Firstly, an 'emotional representations' subscale was designed to assess patients' emotional response to their illness. The SRM proposes that emotional representations are processed in parallel with cognitive representations and may evoke different coping strategies. This component of the model is often neglected and research efforts have mainly focused on the cognitive level. Secondly, an 'illness coherence' subscale was developed to assess the extent to which individuals have a coherent understanding of their illness (Moss-Morris et al. 2002). Moss-Morris et al. (2002) test the application of the IPQ-R across 8 patient populations. Factor analysis identified and confirmed

the 7 dimensions and the sub-scales in the final version were found to show excellent psychometric properties (Moss-Morris et al. 2002).

3.4.5 Illness representations and outcome

A number of studies have investigated the impact of illness perceptions on patients' response to illness.

3.4.5a Functional outcomes

Beliefs about the identity, consequences and control of chronic illness appear to be important predictors of functional response following surgery. In a prospective study Petrie et al. (1996) found patients initial perceptions of the consequences of MI predicted functional recovery including recreational activities and return to work at 3 and 6 months follow up. Orbell et al. (1998) found beliefs regarding the cause and control of osteoarthritis prior to surgery predicted functional activity on follow-up, 9 months later. Perceptions of the identity, consequences and control of CFS and Addisons disease have been associated with functional ability and physical functioning (Moss-Morris et al. 1996, Heijmans and de Ridder 1998, Heijmans 1999).

Scharloo et al. (1998) found beliefs about identity predict physical functioning in patients with psoriasis and beliefs about identity, control and cure account for variance in physical functioning in patients with RA.

3.4.5b Illness management

Illness perceptions also appear predictive of self-management and use of health services. Petrie et al. (1996) and Cooper et al. (1999) found initial beliefs regarding the controllability of MI predicted patient's subsequent attendance for rehabilitation. Hampson et al. (1990) reported that perceptions of diabetes as a serious condition and beliefs in the importance of treatment predicted self-management activities including diet and exercise. Patients with osteoarthritis who believed their illness had more symptoms and was a serious condition made greater use of medical services and engaged in more self-management activities (Hampson et al. 1994). In a year long prospective study of patients with psoriasis, beliefs at baseline (concerning identity, consequences and control) predicted the number of visits made to an outpatient clinic over the subsequent year (Scharloo et al. 2000).

3.4.5c Psychological response

All of the dimensions of illness representation have been associated with the psychological well-being of patients. Beliefs about the identity, cause, manageability, chronicity and seriousness of CFS have been associated with mental health and psychological adjustment to illness (Heijmans and de Ridder 1998, Heijmans 1998 and Moss-Morris et al. 1996). Murphy et al. (1999) found depression in patients with RA to be positively associated with beliefs regarding the identity and consequences of the disease and negatively associated with beliefs about control or cure. Patients who RA initially believed their disease was curable or believed they were personally responsible for disease onset have exhibited increased depression over time (Schiaffino et al. 1998). Orbell et al. (1998) also found beliefs regarding control, consequences and cause of osteoarthritis were associated with levels of depression following surgery. In a prospective longitudinal study Schiaffino et al. (1998) reported that perceived symptom variability was associated with increased depressed mood in patients with multiple sclerosis at 4 month follow up. Scharloo et al. (2000) reported that a great illness identity predicted depression and poor mental health in patients with psoriasis after controlling for the confounding effect of illness severity and duration. Worry about psoriasis has also been associated with beliefs about the seriousness of the condition and emotional cause of the disease, independent of the clinical severity of the condition (Fortune et al. 2000).

These studies provide strong evidence of the role of illness representations in response to chronic illness. A few studies have investigated the impact of both illness representations and coping on response to illness (Heijmans 1998, 1999, Moss-Morris et al. 1996, Scharloo et al. 1998). These studies suggest that illness representations are associated with coping although there is limited evidence for a mediation effect. Illness representations appear stronger predictors of outcome than do coping measures. This may suggest that illness representations have a direct effect on outcome not mediated by coping strategies or that the coping measures used are not adequate to detect an effect.

A number of methodological difficulties have limited the conclusions that can be drawn from these studies. The majority of studies used cross-sectional designs and

correlational analysis. In such studies it is impossible to determine whether individuals view their illness more negatively because they are depressed or vice versa. Causal relationships are difficult to establish. However, a few longitudinal studies have suggested that cognitive representations precede illness outcomes (Petrie et al. 1996, Orbell et al. 1998, Schiaffino et al. 1998, Scharloo et al. 2000). Intervention studies influencing cognitions or coping strategies are needed to provide firm causal evidence. Although some studies have reported illness perceptions to be independent of objective measures of illness severity and disability (Petrie et al. 1996, Scharloo et al. 1998, 2000, Fortune et al. 2000) a number of studies are confounded by illness severity. In this case patients negative illness perceptions and illness outcomes may in fact reflect reality. There is a need where possible to include objective, clinical indices of illness for control purposes. It is however recognised that for some diseases (eg CFS) no objective measures of illness severity exist (Moss-Morris et al. 1996).

3.4.6 Illness perceptions of breast cancer patients

Leventhal's early work on breast cancer patients found that patients held strong perceptions of the identity of breast cancer and described a number of symptoms associated with the disease (Leventhal et al. 1986). Causal attributions for the disease fell into two broad categories of personal habits (diet, stressful life) and personal vulnerability. A number of studies have found patients hold complex multifactorial models in which the cause of breast cancer is attributed to stress, genetics, environment, specific carcinogens, hormones, diet and breast trauma (Buick et al. 1997, Taylor et al. 1984, Stewart et al. 2001). Leventhal et al. (1986) found that patients' representations of breast cancer varied as a reflection of their experience, including variations in type of carcinoma, natural history of the disease and treatment type. Some patients believed the disease to be contained in time and location whereas others perceived the disease as widespread. Some women believed breast cancer to be a chronic, cyclical illness characterised by recurrence whereas other women believed the disease to be acute and short lived. Buick et al. (1997) found that recommended treatment (in their study: either adjuvant radiotherapy or chemotherapy) had a large impact on women's perceptions of breast cancer. Prior to treatment, chemotherapy patients perceived breast cancer as longer lasting and

holding more consequences than radiation patients. Buick et al. (1997) also found that causal beliefs changed over time. In particular beliefs about chance and genetics strengthened during and following treatment.

Buick et al. (1997) assessed the impact of illness perceptions of breast cancer on patients' response to treatment. Illness perceptions were important predictors of psychosocial response to treatment, independent of objective illness severity (tumour size, metastases status, axillary node involvement and type of surgery). Negative beliefs held by patients treated with both radiation and chemotherapy (strong illness identity, self blame, beliefs in long duration and severe consequences and low belief in control/cure) were associated with increased psychological distress and poor coping post-treatment. Taylor et al. (1984) investigated the impact of causal beliefs on adjustment to breast cancer and found no particular causal attribution to be associated with better adjustment. However blaming another individual for the cancer was negatively associated with adjustment. Taylor et al. (1984) also investigated beliefs about control of the disease and found that patients who believed they had control over breast cancer or that others (eg doctors) could control the disease showed greater adjustment. Stewart et al. (2001) investigated breast cancer survivors' perceptions of control over disease recurrence. Women who had remained recurrence-free for at least two years believed recurrence had been prevented by a number of factors including their positive attitude, diet, healthy lifestyle, exercise, stress reduction, prayer, complementary therapies, luck and tamoxifen. This suggested that these women felt a degree of personal control over the prevention of recurrence.

3.4.7 Healthy women's perceptions of breast cancer

Few studies have assessed healthy women's perceptions of breast cancer. The available data suggest women base their beliefs about breast cancer on information available in the media and from family and friends. Most women perceive breast cancer as a very serious illness (Roberts et al. 1984, Payne 1990). Roberts et al. (1984) surveyed 810 healthy women in Scotland to determine their knowledge about breast cancer. Knowledge about the disease was found to be poor with few women describing any other symptom of breast cancer than a lump. Payne et al. (1990)

found women had general knowledge about breast cancer although misconceptions regarding cause and treatment were prevalent. Misconceptions regarding the cause of breast cancer included not wearing a bra, taking vigorous exercise, hitting the breast and having had many sexual partners. Other causal beliefs about the disease fell into 2 main categories: stress (personal affective states and external stressors) and personal and environmental hazards (eg smoking). Buick et al. (1997) compared the beliefs of healthy women with breast cancer patients and reported that healthy women were more likely to endorse self-blame and chance as causes of breast cancer than did patients.

Roberts et al. (1984) found that the majority of healthy women perceived little chance of surviving breast cancer. Although 87% believed that early diagnosis improved chances of survival over 40% of women believed it was not possible to detect the disease in its early stages. Only 31% of women believed it was always or usually possible to cure the disease. In this study 45% of women knew of no other treatment for breast cancer than surgery and only 17% of respondents referred to chemotherapy (Roberts et al. 1984). In contrast, Buick (1997) found healthy women's beliefs regarding the duration and impact of treatment were comparable to those held by patients receiving chemotherapy but not those receiving radiotherapy. In addition, healthy women also tended to overestimate the distressing nature of cancer treatment compared to patients (Buick 1997). Payne (1990) found women to believe in a number of treatments for breast cancer other than surgery. Women in this study reported a variety of 'dietary treatments' (e.g. vitamins and low fat diet) as well as 'psychological treatments' (e.g. learning to relax, and cope with stress). These items appeared to overlap with personal control over breast cancer rather than treatment methods.

Beliefs about breast cancer have been associated with detection behaviour. Payne (1990) reported that women who practiced BSE were more likely to believe in the efficacy of surgical and 'psychological treatments' for breast cancer. Savage and Clarke (1998) conducted interviews designed to elicit older women's (> 45 years) representations of breast cancer and found that women who participated in screening were more likely to cite positive examples regarding the success of breast cancer

treatment in their social circle than non-screeners who were less likely to believe in the efficacy of cancer treatments.

3.4.8 Perceptions of genetic illness

We have little understanding of how awareness of genetic predisposition will affect representations of disease. Information about genetic risk is most likely to influence beliefs about cause and control or cure of illness. Michie et al. (1996) conducted semi structured interviews with individuals at risk of inheriting a gene predisposing them to familial adenomatous polyposis (FAP). This is an inherited form of bowel cancer that shows 100% penetrance. However, individuals were found to hold multifactorial beliefs about the cause of the disease including both genetic and lifestyle factors. Senior et al. (2000) reported an experimental study in which participants were provided with hypothetical test results relating to either heart disease or arthritis. Participants who were informed that the test was genetic were more likely to attribute the illness to genes and less to lifestyle, and to rate both conditions as less preventable, than participants who were not informed of the nature of the test. However this study was conducted on participants with limited experience of the illness. It remains to be seen how knowledge about genetic risk will influence the beliefs of clinical populations with a family history of the disease in question.

Marteau and Senior (1997) argue that a thorough understanding of individuals' representations of genetic illness needs to incorporate not only beliefs about the disease but also beliefs about genes and inheritance. Lay understanding of genetics and beliefs about inheritance within the family and are likely to influence illness representations specifically of the cause and control/cure dimensions.

3.4.9 Perceptions of breast cancer in women at increased risk

Women with a family history of breast cancer have experienced the disease in their family and are likely to have concrete images of the disease and its effects. Chalmers and Thompson (1996) reported that the process of adapting to breast cancer risk was achieved by rehearsing how to cope with personal development of cancer. Illness representations derived from experiences in the family are likely to provide a meaning about ones own risk and expectations about the potential illness experience. However, the variable nature of breast cancer may lead women to develop different

beliefs regarding the identity, timeline, consequences, control/cure and cause of the disease. Women may have observed the effects of different breast cancer types and treatments and may or may not have witnessed recurrence or terminal breast cancer. Within the family, different levels of exposure to breast cancer and its effects, communication about the disease and differences in the personality and coping style of affected relatives may also influence how breast cancer is perceived. Experiences may range from exposure to positive role models, who survived breast cancer and coped well with the disease to more negative experiences in which relatives suffered physical and mental hardship before death. This is demonstrated in the following quotations:

> *"I'm lucky in the fact... that I have... more surviving relatives that have had breast cancer and are fine and that's a big inspiration... You think well its beatable its not a death sentence"* (Appleton, 1999 quote from telephone focus group study of women at increased risk of breast cancer, personal communication).

> *"It would be unpleasant to lose a breast, but dying the horrible death my mother died...I'll never forget it. The three children knew, everyone knew there was a waiting period until she died. The operation was very painful and the scar was hideous. It was difficult to simulate normal appearance. Then she found out she had one year to live. She was in pain all the time. She hated putting her family through it. The end is horrible. You get thin and your hair falls out. The cost is horrible. My father himself paid out $70, 000 plus the insurance. I was going back and forth, bit it was hard for my family to pay for my airfare. I couldn't stand to stay there and watch mother waste away. My mother hated for me to be there and wanted me there at the same time. Our family situation deteriorated."* (Kelly 1983, pg 12)

3.5 SUMMARY

Although studies have shown that women's experience of breast cancer in the family influence their response to genetic risk these studies have largely been a-theoretical and are unable to indicate the causal mechanisms involved. A number of theoretical perspectives have been discussed in this chapter that enable us to see how experiences of cancer in the family may influence psychosocial response to risk and indicate factors amenable to modification. Experiences of breast cancer in the family

have the potential to create bias in interpretation of risk information, contribute to cognitive factors involved in decision-making and impact on illness representations of breast cancer.

Experiences of cancer in the family may create bias in the interpretation of genetic risk information in a number of ways. Firstly, by increasing the availability of images of cancer; secondly, by providing evidence for and expectation of personal risk and thirdly, by enhancing feelings of similarity and resemblance to affected relatives. This bias may be particularly strong when interpreting genetic risk information since lay understanding of genetics and inheritance is often based on resemblance and shared inheritance.

A number of cognitive factors highlighted in the health behaviour change models may be influenced by direct communication about breast cancer and its prevention in the family, observation of affected relatives and by decisions and health-related outcomes of other family member at increased risk. In this way experiences of breast cancer in the family may ultimately influence individuals' decisions about their own risk management (e.g. attending for screening, genetic testing, chemoprevention or prophylactic surgery).

Focusing on the role of cognitive factors (e.g. risk perception) as determinants of psychosocial response to risk neglects emotional aspects of risk. The SRM moves beyond a narrow assessment of perceived probabilities to explore the wider meaning of health threats as perceived by the individual. The model shows how a wide range of concrete experiences, knowledge, memories and social processes are integrated to form both a cognitive and emotional representation of the health threat. The model is able to explain how these components are interrelated in an interactive, dynamic manner and provides a useful framework from which to understand women's psychosocial response to breast cancer risk. This framework may be particularly helpful in understanding variation in levels of distress in women at increased risk of breast cancer.

A body of research has identified and classified dimensions that cover the scope of individual's representations across a range of illnesses. Although the majority of work has been conducted on patients, healthy individuals have also been shown to hold representations of a range of diseases (Bishop and Converse 1986, Bishop et al. 1987). The IPQ has made a major contribution to research in this area by providing a psychometrically sound quantitative measure of illness perceptions that can be applied to most chronic illnesses. It has allowed researchers to investigate associations among dimensions, explore patterns across the dimensions and test the predictive value of single dimensions and combinations of beliefs on outcome variables of interest. Understanding of illness representations in patient samples (including breast cancer patients) has successfully increased understanding of individual differences in patient's functional and emotional response to illness, treatment and illness management. The IPQ has been adapted to assess the perceptions of non-patient samples i.e. spouse of patients (Weinman et al. 1996, Heijmans 1999). However, no work to date has investigated the influence of representations of cancer in healthy individuals at increased genetic risk. There has been little work addressing healthy women's perceptions of breast cancer and there is limited understanding of perceptions of illness associated with genetic predisposition although this is recognised as an important area for research. Applying the SRM to women at increased risk of breast cancer is likely to enhance understanding of psychosocial response to genetic risk.

CHAPTER 4

AIMS, DESIGN AND METHODS

AIMS, DESIGN AND METHODS

4.1 RESEARCH QUESTIONS AND AIMS OF EMPIRICAL WORK

4.1.1 Research questions identified from the literature

The literature reviewed in Chapters 1-3 have identified a number of outstanding research questions that have guided the aims and objectives of the current research. The literature reviewed in Chapter 1 highlighted that the aetiology and maintenance of distress in women at increased risk of breast cancer is poorly understood and understanding of factors that account for variability in psychological adjustment to risk is limited. There is a growing need to understand what factors contribute to increased distress in women at increased risk of breast cancer in order to help identify women who may suffer adjustment difficulties and inform intervention to improve psychological wellbeing in this population.

A number of qualitative studies and anecdotal reports reviewed in Chapter 2 indicated that experience of breast cancer in the family is an important factor that may be linked with poor psychological adjustment to risk. However it is still unclear *what* experiences contribute to distress in women at increased risk breast cancer and the strength of these relationships. Quantitative studied are needed to explore this in more detail. The theoretical perspectives reviewed in Chapter 3 suggested that experience may impact on adjustment to personal risk via cognitive as well as emotional response mechanisms, which are amenable to change. Theoretically driven research is required in order to investigate *how* women's experiences contribute to psychosocial response to risk in order to understand the causal mechanisms involved.

The SRM was described in Chapter 3 and provides a useful theoretical background from which to understand and predict the impact of experience of breast cancer in the family and subsequent illness representations on psychosocial response to genetic risk. The model suggests that awareness of risk status will stimulate the formation and development of illness representations. Women with a family history of breast cancer are likely to develop strong representations of the disease on the basis of their

experiences within their family. These perceptions may provoke anxiety about breast cancer risk and help explain variation in psychosocial response. This model and associated measures have not been applied to at risk population before. The model provides a framework to guide future research and generates a number of research questions including:

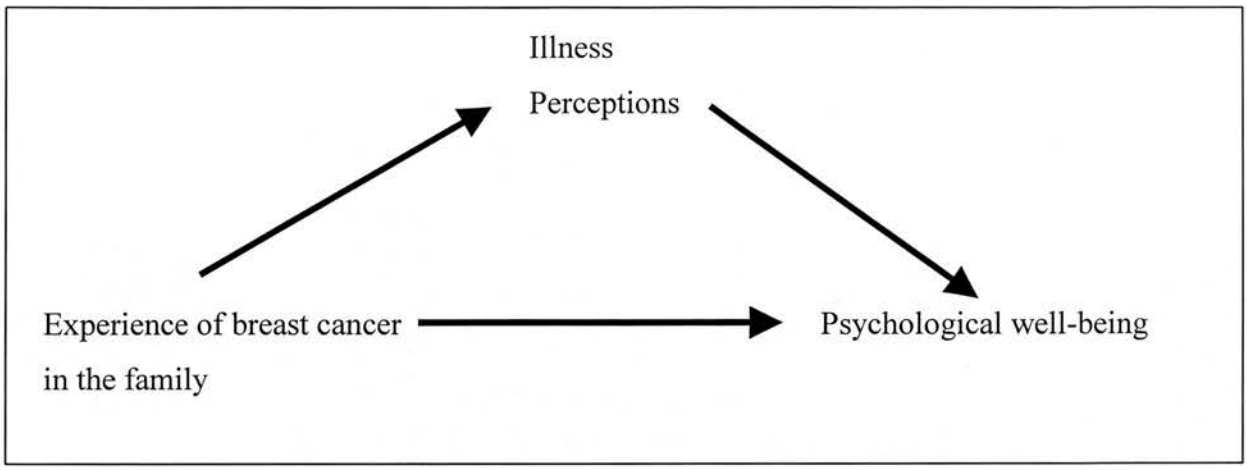
- Can existing measures of illness perceptions be adapted to 'at risk' populations?
- What perceptions of breast cancer do women at increased risk of the disease hold?
- Do women at increased risk of breast cancer hold different illness perceptions to women in the general population?
- What experiences are associated with illness perceptions?
- Are illness perceptions associated with psychological well-being in women at increased risk of breast cancer?

4.1.2 Theoretical model, aims and objectives.

The subsequent work reported in this thesis applied the SRM to the problem of understanding individual differences in women's emotional response to breast cancer risk. The primary aim of the subsequent research was to:

- Test the mediation model (Figure 4.1) that perceptions of breast cancer mediate the impact of experiences of the disease in the family on psychological well-being in women at increased risk of breast cancer.

Figure 4.1- Experience of breast cancer in the family and emotional response to personal risk: A theoretical model.



Secondary objectives of the research included:

1. To evaluate the use of the IPQ-R to assess perceptions of breast cancer in women at increased risk and women in the general population.
2. To examine associations between experience of breast cancer and levels of general and cancer specific distress in women at increased risk of breast cancer and women in the general population.
3. To examine associations between experience of breast cancer and perceptions of the disease in women at increased risk of breast cancer and women in the general population.
4. To examine associations between perceptions of breast cancer and levels of general and cancer specific distress in women at increased risk of breast cancer and women in the general population.

4.1.3 Hypotheses

This theoretical model suggests a number of hypotheses that will be tested throughout this thesis. The main hypotheses dealt with in subsequent chapters are outlined below.

4.1.3a Experience and distress (Chapter 7)

H1. Women with a significant family history of breast cancer will show higher levels of cancer specific distress than women without any experience of breast cancer.

H2. Experiences of breast cancer in the family will be associated with levels of general and cancer specific distress in women at increased risk.

H3. Experiences of breast cancer reported by women in the general population will be associated with levels cancer specific distress in this sample.

4.1.3b Experience and illness perceptions (Chapter 8)

H4. Women with a significant family history of breast cancer will hold more negative perceptions of the disease (higher scores on the identity, timeline acute, consequences and emotional representations subscales of the IPQ-R) than women without any experience of breast cancer.

H5. Experiences of breast cancer in the family will be associated with perceptions of the disease in women at increased risk.

H6. Experiences of breast cancer in family or friends reported by women in the general population sample will be associated with more negative perceptions of the disease (higher scores on the identity, timeline acute, consequences and emotional representations subscales of the IPQ-R).

4.1.3c Illness perceptions and distress (Chapter 9)

H7. Perceptions of breast cancer will be significantly associated with levels of general and cancer specific distress in women with a significant family history of breast cancer.

4.1.3d The mediation model (Chapter 10)

H8. The impact of experience of breast cancer on levels of general and cancer specific distress will be mediated by perceptions of breast cancer.

4.1.4 Analysis Plan

The hypotheses outlined above will be tested in the context of a cross-sectional comparison of women at increased risk of breast cancer and women in the general

population. Analysis will be conducted at two levels. Firstly, differences between the samples will be examined (H1 and H4). Secondly associations between experience of breast cancer and levels of distress (H2-H3); experience of breast cancer and illness perceptions (H5-H6); as well as illness perceptions and levels of distress (H7) will be further investigated in each sample. This analysis will be supplemented by additional multivariate analysis in the increased risk sample in order to help identify the best predictors of distress. The analysis will identify potential mediation models that will be examined in the final results chapter (Chapter 10). An outline analysis plan in provided in Table 4.1 Details of specific statistical techniques are provided where appropriate.

Table 4.1- Summary of analysis plan.

Chapter	Results	Analysis
Chapter 5	Sample	• Descriptive statistics • Comparison of respondents and non respondents • Comparison of respondents in each sample
Chapter 6	Evaluation of measures of experience and illness perceptions	• Descriptive statistics • Scaling analysis • Reliability analysis • Correlational analysis • Cluster analysis
Chapter 7	Experience of breast cancer and distress	• Comparisons of samples • Associations between experience and distress within each sample
Chapter 8	Experience of breast cancer and illness perceptions	• Comparisons of samples • Associations between experience and distress within each sample
Chapter 9	Illness perceptions and distress	• Associations between illness perceptions and distress in each sample • Predicting distress in the increased risk sample.
Chapter 10	The mediation model	• Multiple regression analysis to test mediation

4.2 DESIGN

A cross sectional postal questionnaire study comparing women at increased risk of breast cancer (Sample A) and women in the general population with (Sample B) and without experience of breast cancer (Sample C) was used. A sub-sample of the increased risk sample was also followed up at 3-months with an additional questionnaire to obtain test-retest reliability data.

4.3 PARTICIPANTS

4.3.1 Sources of participants

4.3.1a Increased risk sample (Sample A)

Women on the database of the South East Scotland Familial Breast Cancer Clinic, who had been assessed as being at least 'moderate' increased risk of breast cancer and were maintained on surveillance (i.e. regular mammogram and clinical examination). At the time of data collection only women who had received genetic counselling and had met criteria for being at increased risk of breast cancer (see Figure 1.1) were recorded on the database. The judgement of risk status had been made by the clinician following genetic counselling session. At time of data collection objective risk status was not collated on this database.

4.3.1b General population sample (Samples B&C)

Women were identified from the Community Health Index at Lothian Health. This is a register of all individuals registered with GPs in the Lothian region.

4.3.2 Inclusion criteria

4.3.2a Increased risk sample (Sample A)

Any woman at increased risk of breast cancer due to a family history of the disease.

4.3.2b General population sample (Sample B&C)

Women were selected on the basis of their age and postcode sector (eg EH10 4) to be comparable to the increased risk sample. This sample was divided into women with

experience of breast cancer in their social environment (Sample B) and women without any experience of breast cancer (Sample C).

4.3.3 Exclusion criteria

It was important that the increased risk sample was as homogeneous as possible since variability in the sample may affect levels of distress or illness perceptions. Therefore, women on the database of the South East Scotland Familial Breast Cancer Clinic were excluded prior to selection if they had had preventative surgery, chemo-prevention or genetic testing. Women with an ovarian family history were also excluded. Women were also excluded from the database if they had taken part in any other psychosocial research project in the last six months. Women in both samples were excluded for the following reasons:

- Previous diagnosis of cancer in the past
- Currently undergoing investigation for cancer
- Currently suffering from another serious illness
- Currently suffering from alcoholism, schizophrenia or organic brain damage

4.4 MEASURES

4.4.1 Background demographics

Postcode sector was converted into a measure of social deprivation using the Carstairs Index (Carstairs and Morris 1991). This is a measure of social deprivation based on 4 criteria: overcrowding, male unemployment, low social class and proportion of individuals within private households without a car. The scores are collapsed into two scales a 5 point scale (Depcap 5) and a 7 point scale (Depcap 7). High scores on both scales represent higher levels of deprivation (Carstairs and Morris 1991).

In both samples participants were asked to report their marital status, educational level and ethnic group. Women were also asked if they had any children, and if so the age and gender of each child.

Women were also asked their perception of risk with the following two questions:

How likely do you feel it is that you will develop breast cancer in your lifetime?

Very Unlikely	Unlikely	Likely	Very likely	Extremely likely
1	2	3	4	5

Do you think that your risk of ever developing breast cancer is:

a. Lower than the general population ☐
b. The same as the general population ☐
c. Slightly higher than the general population ☐
d. Much higher than the general population ☐

4.4.2 Experience

4.4.2a Increased risk sample (Sample A)

The experience questionnaire is described in Chapter 6. A copy of the instrument in given Appendix II (page A-4)

4.4.2b General population sample (Sample B&C)

Participants in the general population sample were asked the following questions regarding their experiences of breast cancer for control purposes.

- Have any of your relatives ever suffered from breast cancer? (yes/no)
- If yes, please list what relatives have suffered from breast cancer (eg mother, sister etc)
- Have any of your family or friends suffered from breast cancer recently? (ie in the past 12 months) (yes/no).
- Does your work bring you into contact with cancer patients on a regular basis? (yes/no, if yes please give details)
- Do you have any other experiences of breast cancer you would like to tell us about? (open ended).

Response to these items were used to create sub-samples. Participants who reported any experiences of breast cancer were allocated to Sample B (general population sample with experience of breast cancer) and those who did not report any

experience of breast cancer were allocated to Sample C (general population sample without any experience of breast cancer).

4.4.3 Illness perceptions

The IPQ-R (Weinman et al. 1996, Moss-Morris et al. 2002) described in Chapter 4 (section 3.4.4, page 84) was adapted to measure illness perceptions of breast cancer in both samples. This measure included assessments a number of dimensions of illness perceptions: identity, timeline acute/chronic, consequences, timeline cyclical, personal control, treatment control, illness coherence, emotional representations, causes. The adaptation of the questionnaire, description of subscales, scoring and details of reliability shall be covered in detail in Chapter 6.

4.4.4 Distress

Measures of both general and cancer specific distress were included in this study (see Chapter 1, section 1.4.1, page 50, for a description of measures of general and cancer specific distress in women with a family history of breast cancer).

4.4.4a General distress

The 30-item version of the General Health Questionnaire (GHQ-30) was used to assess general distress. This is a self-administered screening test aimed at detecting psychiatric disorder in the community and non-psychiatric clinical settings. Participants respond to thirty statements about their general health over the past few weeks on a 4 point scale. The GHQ-30 was scored using the 'alternative' scoring system (0, 0, 1, 1) to determine the prevalence of psychopathology. In this method responses are summed to achieve a total score. Using this method a score over 5 is taken as the cut-off point to indicate levels of distress that warrant further clinical assessment ('caseness') (Goldberg and Williams 1988). Responses were also scored using the 'Likert' scoring system (0, 1, 2, 3) in order to investigate individual differences in levels of distress. There are numerous studies on the measures internal reliability (mean 0.87), test-retest reliability (ranging from 0.51-0.9) and validity (correlation coefficients of 0.45-0.77 when compared to interview measures of morbidity) (Goldberg and Williams 1988). The scale has been used previously to assess levels of distress pre and post genetic counselling at the Edinburgh familial breast cancer clinic (Cull et al. 1998, 1999).

4.4.4b Cancer specific distress

Two measures of cancer specific distress were used.

4.4.4b(i) Cancer worry scale

The cancer worry scale was used to assess anxiety specific to breast cancer (Watson et al. 1998). This is a 6 item self-administered scale assessing frequency of breast cancer related worries and the degree to which these worries affect mood and daily activities. Each question is scored from 1-4 and summed for a total score. The scale has been used within populations at increased risk of cancer although there is limited published data on its psychometric properties. Brain et al. (1999) report an alpha reliability coefficient of .86 when administered to a sample of women prior to genetic counselling.

4.4.4b(ii) The Impact of Event scale

The Impact of Event scale was used to determine levels of intrusive and avoidant thoughts about breast cancer. The measure was originally developed to assess subjective levels of distress associated with a specific traumatic event (Horowitz et al. 1979). It is a 15 item scale that measures levels of intrusive and avoidant thoughts occurring in the past week. Seven questions refer to the frequency of intrusive thoughts and eight to avoidance. Participants respond to each statement on a 4-point scale ranging from 'not at all' to 'often'. When scored the responses are weighted 0, (not at all), 1 (rarely), 3 (sometimes) and 5 (often). Original split half reliability reported for the scale was r= .86. Cronbach's alpha values were reported as 0.78 for the intrusion subscales and 0.82 for the avoidance subscale (Horowitz et al. 1979). The reliability and validity of the scale have been confirmed in the general population (Briere and Elliott 1998). The scale has recently been modified to determine levels of cancer specific distress associated with risk of breast cancer (Kash et al. 1992). It shows acceptable reliability (split half reliability: r= .86; test-retest reliability: r=.87 intrusion; r= .89 avoidance; Cronbach's alpha reliability coefficient: 0.91 (Kash et al. 1992)) and has been widely used within populations at increased risk (e.g. Lloyd et al. 1996, Gagnon et al. 1996, Watson et al. 1999, Lerman et al. 1994a, Valdimarsdottri et al. 1995, Zakowski et al. 1997, Thewes et al. 2001).

Lloyd et al. (1996) included an opt out box for women who had not thought about breast cancer in the past week (Lloyd et al. 1996). This version of the measure was used in this study because a large proportion of the general population sample were expected not have thought about breast cancer in the past week. A recent psychometric assessment of the scale in women at increased risk of breast cancer respondents criticised the measure for not including a response options for those who had not thought about breast cancer (Thewes et al. 2001).

4.4.5 Follow up questionnaire measures

The follow up questionnaire was administered to a sub-sample of the increased risk sample and contained the same measures of distress and illness perceptions outlined above. The experience items were not repeated, as test-retest data were not required. However, it was necessary to control for women's experiences *between* questionnaires that may have influenced anxiety or perceptions of breast cancer. Women were asked to report any experiences that may have influenced their thoughts and feelings about breast cancer. Prompts were given for experiences which have been reported to influence feelings about breast cancer risk such as attending for screening and media reports (Appleton et al. 2000) as well as general experiences expected to influence levels of distress or perceptions of breast cancer. The items included in the questionnaire were:

- Since you last filled in this questionnaire about 3 months ago have you attended the Ardmillan Familial Breast Cancer Clinic for an annual check up? (Yes/No)
- Since you last filled in this questionnaire, can you think of any experiences that may have changed your thoughts and feelings about breast cancer? (Yes/No)
- If yes, please let us know about these experiences by ticking the box(s) and describing your experiences:
    - Events at the clinic
    - Family experiences
    - Experiences of friends
    - Experiences at work
    - Media reports
    - Other

4.5 PROCEDURE

The database from the SE Scotland Familial Breast Cancer Clinic was imported into SPSS and a random sample of 200 women selected using the random number selection. The following information was then obtained for this sample:

- Date of birth
- Address including postcode
- GP name and address.

This information was used to select a comparable sample of women in the general population. For each woman at increased risk 2 women in the general population with the same postcode region and date of birth within 1 year were selected (n=400). Following a low response rate in the general population another match for each woman at increased risk was obtained at a later date (n=200). For 8 of the women at increased risk only 2 appropriate matches could be identified and hence only 592 women in the general population were selected.

The women's GP's were contacted in order to ensure the exclusion criteria (see 4.3.3, page 104). GPs were provided with an information sheet about the study and asked to contact the researcher by email, telephone or fax within the next fortnight if they considered the patient was not suitable for the study because of the exclusion criteria or for any other reason. The GPs were informed that if they had not contacted the researcher within 2 weeks it would be assumed that the patient was eligible to participate in the study.

Participants who were eligible for the study were sent an information sheet and consent form and invited to take part in the study. Women were asked to indicate on the consent form if they wished to participate in the research project or not and to return the form in the freepost envelope provided. Women who consented were sent a questionnaire pack and a freepost envelope for its return. If the questionnaire was not returned within 3 weeks another copy of the questionnaire was sent with a reminder letter. In the initial general population sample (n= 400) women who did not

return their consent form were sent a reminder letter and another consent form in an attempt to increase the sample size.

In order to obtain test-retest reliability data in the increased risk sample a second questionnaire was sent to a subsample of the increased risk sample (participants who had returned the first questionnaire without prompting) 3 months after they were sent the initial questionnaire.

4.6 ETHICAL CONSIDERATIONS

Ethical approval for this study was obtained from Lothian Research Ethics Committee. The SE Familial Breast Cancer Clinic is a research led clinic and women who attend the clinic are aware that they may be approached for clinical trials (e.g. IBIS, see Chapter 1, section 1.2.5, page37) or other research. A number of research projects were being conducted at the time of this study. It was essential therefore that the research aims be addressed as economically as possible, ideally within the context of a single study. In order to ensure women on the database were not over-exploited for research studies women who had participated in any other psychosocial research project in the last six months were excluded. This also guaranteed that women selected had not participated in the previous pilot work concerning development of the experience questionnaire (see Chapter 6, Part 2, sections 6.3-6.8). Women were provided with an information sheet regarding the study that clearly stated that their decision to participate in the research or withdraw at any stage would not affect the services they would receive now or in the future.

Initially women's GP's were contacted in order to check the exclusion criteria (see 4.3.3, page 104) and to determine any additional concerns about approaching their patients. An 'opt out' response format was used in order to reduce the amount of time the GP needed to spend on this task.

For the general population sample it was necessary to adhere to strict confidentiality guidelines concerning information stored at Lothian Health Board on the Community Health Index (CHI). Information stored on this database is confidential and direct access is only allowed for the purposes of providing services to patients. However,

Lothian Health Board occasionally allow information on the CHI to be used for ethically approved medical research studies. A research officer from Lothian Health Board selected the sample based on the criteria provided and made contact with the GP and subsequently the patient, if the inclusion criteria were met. Lothian Health Board included an additional information sheet to both the GP and patient to explain the use of the CHI and to ensure that no personal information about the patient would be available for research purposes unless the patient consented to the study. Lothian Health Board provided a list of anonymous date of births and postcode regions of the initial sample prior to contact with the GP as well as the number of participants who were excluded by the GP (and reasons where provided). Contact details were only provided by the patient, if consenting to the study.

Confidentiality was maintained during data management. Each participant was allocated a unique study code and this code was the only identifier that appeared on any on the participants data gathered during the study. Raw data was stored in locked filing cabinets and data stored on computer was password protected.

Support was available if women found the research distressing. Contact details of a nurse at the Familial Breast Cancer Clinic were provided on both the information sheet and questionnaire pack if women required further information about the study or if the study raised any issues of concern. Contact details of the principal researcher were also provided. Resources of the Department of Clinical Psychology were also available for psychological support in the unlikely event that the questionnaire caused severe distress or worry. Women's GP and the Familial Breast Cancer Clinic were notified if any participant scored above the cut-off point on the GHQ-30.

4.7 SAMPLE SIZE

A minimum of 100 women in each sample was required in order to achieve statistical power for multivariate analysis of associations between measures of experience, illness representations and distress. A sample size of 100 will be able to detect r-squared values (percentage of variance of the dependent variable accounted for by the model) of 10-15% in multiple regression models with 80% power (Hair et al. 1995).

Sample size was also required in order to ensure a sufficient sample size to detect differences in illness representations between women with different levels of distress However, no published data were available on the differences between groups using the IPQ-R hence power calculations using this measure could not be computed.

A response rate of 50% was expected, taking into account low response rates to questionnaires (Altman 1997). A sample of 200 women at increased risk was therefore selected. As no information about the experiences of women in the general population sample could be ascertained prior to the questionnaire a larger sample was anticipated as being necessary in order to achieve a sample of 100 controls with no experience of breast cancer in family or friends. Initially, 400 women were selected from the general population. Due to a low response rate a further 192 were approached bringing the full sample size to 592. Across both samples the overall response rate in the study was 50.3%.

CHAPTER 5
DETAILS OF SAMPLES

DETAILS OF SAMPLES

5.1 EXCLUDED PARTICIPANTS

5.1.1 Increased risk sample (Sample A)

From the 200 women identified from the database, sixteen were excluded by the GP prior to the study. Reasons for exclusions are provided in Table 5.1. A sample of 184 women at increased risk was therefore invited to participate in the study.

Table 5.1- Participants excluded from the increased risk sample by the GP.

Exclusion reason	Number of participants excluded
Not registered at GP	8
Depression	2
Anxiety and depression	1
Previous cervical cancer diagnosis	1
Serious illness	1
Previous diagnosis of endometrial cancer	1
Currently under investigation for cancer	1
No reason given	1
Total excluded	16
Sample size remaining	184

5.1.2 General population sample (Sample B&C)

Thirty women in the first general population sample (n=400) were excluded by the GP (please see Table 5.2). In the second general population sample (n=192) an additional 15 women were excluded by the GP and were subsequently replaced (please see Table 5.3). A total of 562 women in the general population were therefore invited to take part in the study.

Table 5.2- Participants excluded by the GP from the first general population sample selection.

Exclusion reason	Number of participants excluded
No reason given	12
Not on GP list	5
Moved from area	4
Currently suffering from cancer	3
Participant is male	1
Currently suffering from another serious illness	1
Emotionally unsuitable	1
Limited IQ	1
Downs syndrome	1
Pregnant	1
Total excluded: Sample size remaining:	30 370

Table 5.3- Participants excluded by the GP from the second general population sample selection.

Exclusion reason	Number of participants excluded
No reason given	10
Surgery to busy to check records	3
Disabled	1
Deceased	1
Total excluded Total replaced	15 15

5.2 RESPONSE RATES

Response rates were calculated from the number of individuals who were deemed eligible for the study (ie not excluded by their GP) who were sent an information sheet and consent form. The proportion of women who returned their questionnaire before and after reminders is reported.

5.2.1 Increased risk sample (Sample A)

Of the 184 women who were deemed eligible for the study, 128 (69.6%) consented to take part. Fourteen women stated that they did not want to participate in the study and 42 did return the consent form. It was presumed these women did not want to participate in the study.

Ninety-nine women returned the questionnaire before reminders (53.8%). A further 18 returned the questionnaire after the reminder giving a response rate of 63.6% (n=117).

5.2.1a Follow up sample

The 99 women in the increased risk sample who returned their questionnaire without prompting were sent a follow up questionnaire. Seventy-four (74.8%) returned a completed questionnaire.

5.2.2 General population sample (Sample B&C)

In the first general population sample (n=370) 145 (39.2%) women consented to the study, 21 did not consent and 204 did not return to the consent form. A subsequent form was sent to those who did not respond and 55 women returned this form stating that they would consent to be study. In the second general population sample (n=192), 77 women consented to the study.

Overall, 562 women were deemed eligible for the study and 277 consented to take part. Two hundred and twenty seven questions were returned before a reminder and 31 received following a reminder. The overall response rate was 45.9% (n= 258). Two respondents had reported on their questionnaire that they had suffered from cancer in the past. As this was an exclusion criterion (see 4.3.3, page 104) these

women were excluded at this stage. The number of women in the general population sample was therefore reduced to 256 (45.5%).

5.2.2a General population sample without experience of breast cancer (Sample C)

One hundred women (39.1%) in the general population did not reported any experiences of breast cancer in family, friends or at work. This sub-sample of the general population formed the control sample in subsequent analyses.

5.3 DESCRIPTION AND COMPARISON OF SAMPLES

Figures 5.1 and 5.2 provides a summary of each samples and response rate throughout the study.

Figure 5.1- Increased risk sample (A): Outline and response rate

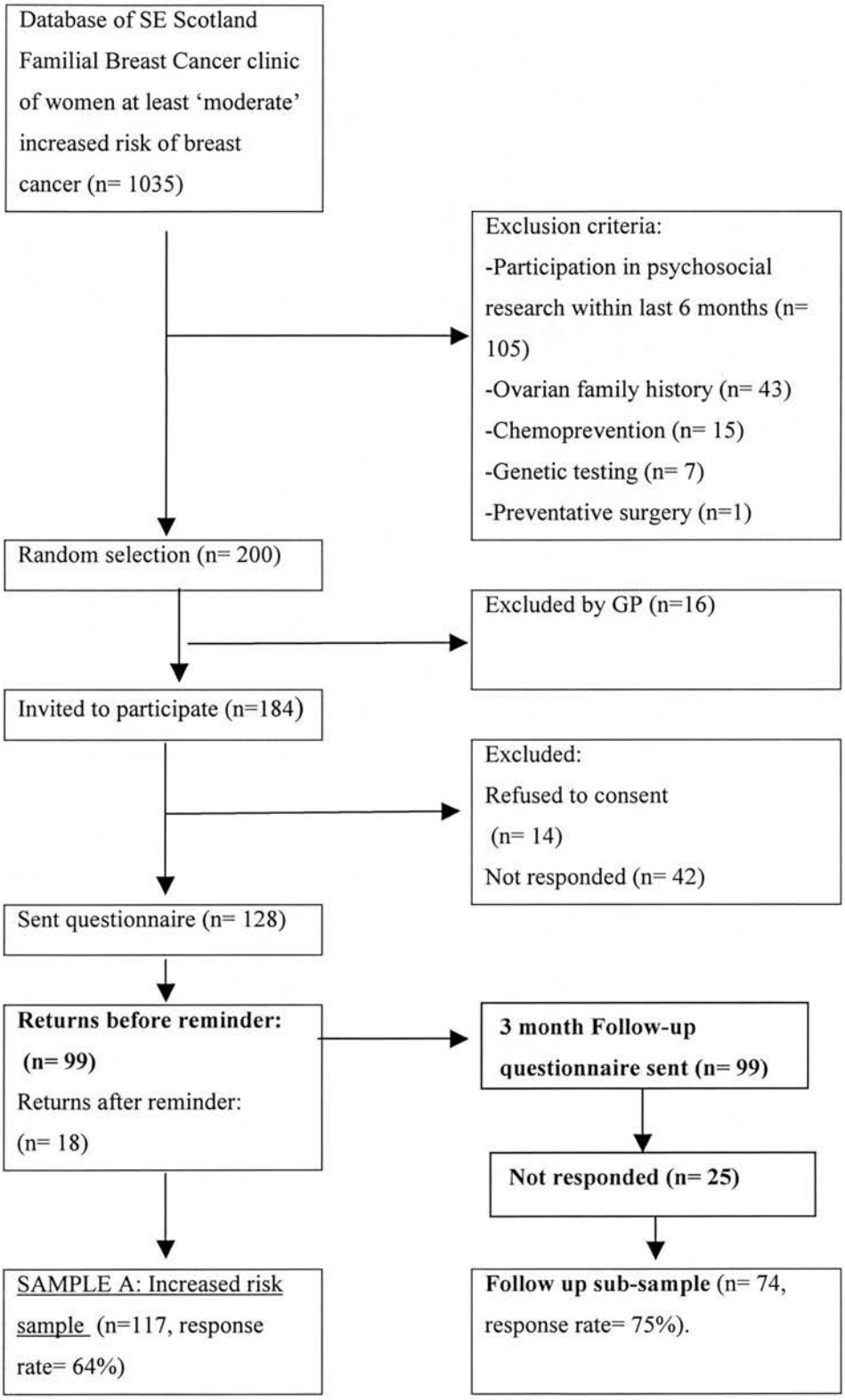
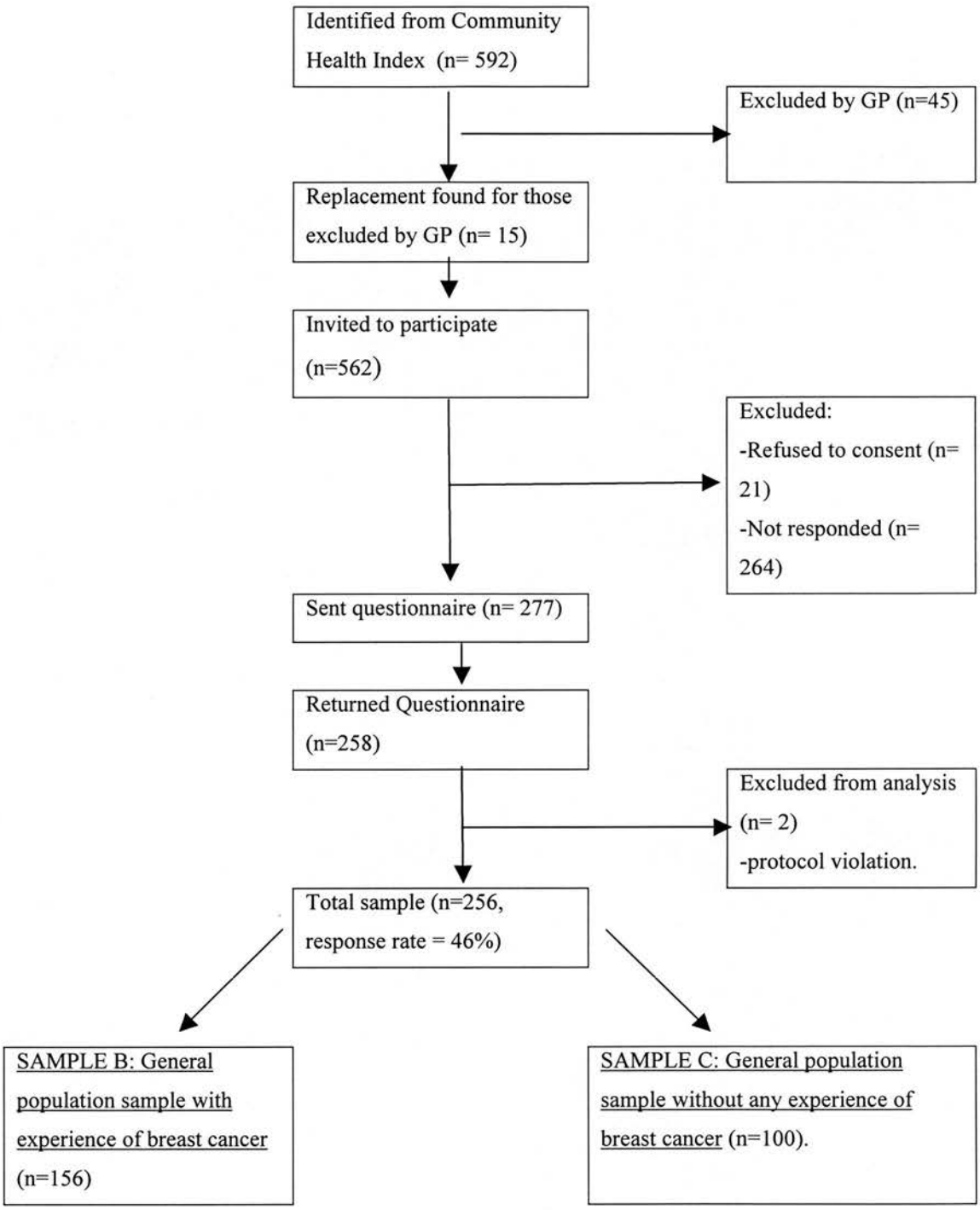


Figure 5.2- General population sample (B&C): Outline and response rate



5.3.1 Comparisons of initial samples

Women selected for the increased risk (n= 200) and general population (n= 592) samples were compared on age and Carstairs Deprivation Score. There were no significant differences in age (t= .02, df= 790, p= 0.98), Depcap 5 (t= .26, df = 790, p= 0.80) or Depcap 7 (t = .83, df= 790, p= 0.41) between the increased risk sample and the full general population sample. Mean values for each sample are provided in Table 5.4

Table 5.4 - Age and Carstairs Deprivation Scores for each sample

Sample	n	Age Mean (sd)	Range	Depcap 5 Mean (sd)	Depcap 7 Mean (sd)
Increased risk (A)	200	39.8 (7.39)	23-59	2.6 (1.31)	3.3 (1.39)
General population (B&C)	592	39.9 (7.33)	23-59	2.6 (1.26)	3.4 (1.37)

5.3.2 Comparisons of respondents and non-respondents in each sample

5.3.2a Increased risk sample (A)

There was no significant difference in age or levels of social deprivation between respondents and non-respondents ($p<0.05$) (Table 5.5). Non-respondents showed a trend to higher levels of social deprivation than respondents on Depcap 5($p<0.1$).

Table 5.5- Differences between respondents and non-respondents in the increased risk sample (Sample A)

	Respondents	**Non respondents**	**df**	**t**	**p**
N	117	83			
Age: Range	23-56	23-59			
Mean (sd)	40.4 (7.09)	39.4 (7.60)	198	.94	0.35
Depcap 5: Mean (sd)	2.5 (1.23)	2.8 (1.40)	198	1.80	.07
Depcap 7: Mean (sd)	3.2 (1.31)	3.4 (1.51)	198	1.34	0.18

5.3.2a(i) Follow-up sample

There were no differences in age or risk perception between participants who were sent a follow up questionnaire and the rest of the increased risk sample ($p<0.05$).

There were also no differences in age or risk perception between participants who returned a follow-up questionnaire and those who did not ($p<0.05$).

5.3.2b General population sample (B&C)

There were no significant differences between respondents and non-respondents on age or the Carstairs deprivation scores ($p<0.05$). A trend on Depcap 7 suggested that the non-respondents in this sample also showed greater social deprivation than respondents (Table 5.6).

Table 5.6- Differences between respondents and non-respondents in the general population sample

	Respondents	**Non respondents**	**df**	**t**	**p**
N	256	334			
Age: Range	25-59	23-59			
Mean (sd)	40.3 (6.99)	39.5 (7.57)	588	1.37	0.17
Dep 5: Mean (sd)	2.5 (1.27)	2.7 (1.27)	588	1.55	0.12
Dep7: Mean (sd)	3.2 (1.34)	3.5 (1.40)	588	1.95	0.052

5.3.3 Comparisons of respondents in each sample

Respondents in the two samples showed comparable socio-demographic characteristics. These are summarised in Table 5.7. There was no difference in age ($t= .55$, $df= 371$, $p= 0.58$); education level ($t=.51$, $df= 368$, $p= 0.61$); marital status or maternity between the general population sample and the increased risk sample (chi-square= .502, df= 1, p= 0.48; chi-square= 440, df= 1, p= 0.51).

Table 5.7 Summary of socio-demographic characteristics for respondents in each sample.

Socio-demographic characteristic	Increased risk sample (A)	General population sample (B&C)
Mean age	40.4	40.8
% Reporting education/training after age 18	45%	48%
% Married or living with a partner	88%	79%
% With at least one child	71%	73%

As expected the samples differed significantly in their perception of personal breast cancer risk. These differences are summarised in Table 5.8 and 5.9. Participants in the increased risk sample (A) were significantly more likely to believe they were likely to develop breast cancer in their lifetime than women in the general population sample (B&C) ($t= 7.83$, $df= 366$, $p<0.001$). Seventy-seven percent of women in the increased risk sample believed it was 'likely', 'very likely' or 'extremely likely' that they would develop breast cancer compared to 38.3% of the general population sample.

Table 5.8- Differences in risk perception (1) between the samples.

How likely do you feel it is that you will develop breast cancer in your lifetime?	Increased risk sample (A). n, (%)	General population sample (B&C). n, (%)
Very unlikely	1 (0.95%)	11 (4.3%)
Unlikely	25 (21.4%)	143 (55.9%)
Likely	70 (59.8%)	89 (34.8%)
Very likely	16 (13.7%)	8 (3.1%)
Extremely likely	4 (3.4%)	1 (0.4%)

The vast majority of women at increased risk of breast cancer (sample A) perceived their risk as slightly or much higher than the 'general population risk of breast cancer' (92.3%). The majority of the general population sample (B&C) perceived their risk as 'the same as the general population' ($t= 19.34$, $df 371$, $p<0.001$).

Table 5.9- Differences in risk perception (2) between the samples.

Do you think that your risk of ever developing breast cancer is:	Increased risk sample (A). n (%)	General population sample (B&C) n (%)
Lower than the general population	0	19 (7.4%)
The same as the general population	9 (7.7%)	201 (78.5%)
Slightly higher than the general population	86 (73.5%)	35 (13.7%)
Much higher than the general population	22 (18.8)	1 (0.4%)

5.3.3a Women in the general population sample without experience of breast cancer (Sample C)

The control sample was the subset of the general population sample who had no experience of breast cancer (see Figure 5.2). There was no significant difference

between the control sample and increased risk sample in age (t= 1.19, df= 215, p= 0.24), education level (t= .460, df= 212, p= 0.65), marital status (chi-square= .018, df= 1, p= 0.89) or maternity (chi-square 2.37, df= 1, p= 0.12). As expected women in the increased risk sample scored significantly higher perceptions of breast cancer risk on both measures than women in the control sample. Women in the increased risk sample more likely to believe they will develop breast cancer in their lifetime (t= 7.48, df= 211, p<0.001) than the control sample. Eighty-seven (87%) of women in the control sample believed their risk of breast cancer was the same as the general population and 67% believed they were 'very unlikely' or 'unlikely' to develop breast cancer in their lifetime.

5.4 SUMMARY AND DISCUSSION

The response rate in the increased risk sample was good but as expected recruitment was much lower in the general population sample. A second phase was required to achieve the specified sample size. The difference in response rate most probably reflects greater interest and relevance of the study for women at increased risk of breast cancer compared to women in the general population.

The general population sample was selected on the basis of information concerning the age and postcode region of the increased risk sample and the initial samples were therefore of comparable age and social deprivation level. No strong bias was found between respondents and non-respondent in each sample although there was a trend in each sample for non-respondents to show greater social deprivation. No differences were found between respondents in each sample on any of the background demographic factors. The increased risk sample was comparable to both the general population sample and the control sample in age, education, marital status and maternity. As expected women in the increased risk sample reported higher subjective risk perceptions than women in the general population sample.

In the general population sample two women had slipped through the exclusion criteria. They reported on their questionnaire that they had developed cancer in the past. These women should have been identified and excluded at the GP stage. This raises the possibility that 2 weeks was insufficient time for GPs to fully check all

exclusion criteria. An 'opt in' rather than 'opt out' approach at the GP stage may have been more thorough. However, this would need to be balanced with the extra requests on GPs for their time.

CHAPTER 6

DEVELOPMENT AND EVALUATION OF MEASURES

DEVELOPMENT AND EVALUATION OF MEASURES

6.1 BACKGROUND: PSYCHOMETRIC EVALUATION OF MEASURES.

Accurate measurement is a fundamental element of scientific research and essential for confident application of research findings. There are a number of criteria to determine if a measure of internal psychological stimuli is psychometrically sound and likely to provide useful information. Johnston et al. (1995) identify these criteria as: applicability; acceptability; readability; sensitivity; specificity; reliability and validity. Measures must be appropriate and comprehensible to the population in question and be sensitive enough to detect differences across individuals or changes over time. The scores obtained on the measure should be reliable and not change unless there has been some change in the phenomenon under assessment. There are two main types of reliability for self-report instruments. Firstly, internal consistency indicates if one part of the measure gives similar scores to other parts of the measure. A common statistic used to assess internal consistency is Cronbach's alpha. This statistic represents a summary of the overall correlation between items. A value of 0.6 is regarded as acceptable for scales in the process of development whereas a value of 0.7-0.8 is deemed as the acceptable lower limit for established tests (Johnston et al. 1995, Kline 1998). Secondly, stability refers to the reproducibility of the measure over time. This is often assessed by administering the measure to the same participants at two points in time and calculating the correlation coefficient between the scores obtained (test-retest reliability). A correlation coefficient value of 0.5 is regarded as reasonable stability (Streiner and Norman 1991). A measure must also prove valid (measure what it is designed to measure). There are a number of aspects of validity: face validity (degree to which items appear to resemble the construct they are designed to assess); content validity (degree to which the measure covers all relevant domains); concurrent validity (associations with other measures designed to assess the same or similar construct); construct validity (degree to which items assess the construct they are designed to sometimes assessed with factor analysis); criterion validity (ability of the measure to discriminate between groups hypothesised to differ on the construct in question); predictive validity (extent to which scores are predictive of future outcomes).

Measures of the three main constructs in this thesis (experience, distress and illness perceptions) required psychometric evaluation to ensure they were adequate for use in subsequent analysis (see Table 4.1, page 102). Published research has demonstrated adequate psychometric properties of distress measures in women at increased risk of breast cancer but these needed to be examined in the general population sample. At the time of this study quantitative measures of experience were not available in the literature and measures of illness perceptions had not been evaluated in healthy populations. The purpose of this study was not to develop new measures but to examine the utility of adapting existing measures required to test the primary aim of the thesis (see Figure 4.1, page 100).

Prior to this research work had already begun to develop a questionnaire to assess important aspects of experience in women with a family history of breast cancer. Prior to the researcher joining the psychosocial research group a quantitative questionnaire addressing experience of breast cancer in the family had been developed based on a limited review of the literature, clinical observation and interviews with women attending the familial breast cancer clinic in Edinburgh. This had been administered to women attending genetic counselling sessions and following analysis of responses a shortened version of the questionnaire had been derived (Cull, personal communication).

A theoretically derived measure of illness perceptions (IPQ) was available in the literature (Weinman et al. 1996) and had recently been revised (IPQ-R) (Moss-Morris et al. 2002). However, this measure had not been evaluated for use in healthy samples. Adapting this measure would allow comparison with research findings in patient populations and also provide evidence regarding the potential application of the measure in healthy samples. Three sets of analyses are reported in this chapter. Part 1 outlines the reliability details of the cancer specific distress measures in this study. Part 2 refers to the adaptation and assessment of experience of breast cancer in the family. Part 3 reports the adaptation and evaluation of the IPQ-R to assess healthy women's perceptions of breast cancer.

PART 1

CANCER SPECIFIC DISTRESS MEASURES

6.2 RELIABILITY

6.2.1 Cancer worry scale

In the increased risk sample (A) the mean score on the cancer worry scale was 10.2 (sd= 2.58, n= 116). In the general population sample (B&C) the mean score was 9.1 (sd= 2.27, n= 253). Cronbach's alpha was 0.8 in the increased risk sample (A) and 0.78 in the general population sample (B&C). Test-retest reliability in the increased risk sample was r= .75 (n=73, p< 0.001).

6.2.2 Impact of Event scale

The mean scores on the Impact of Event scale are provided in Table 6.1. Internal consistency of the measure was acceptable in the increased risk sample (A) (Cronbach's alpha = .92 (total), .87 (intrusion), .87 (avoidance)) and the general population sample (B&C) (Cronbach's alpha = .93 (total), .89 (intrusion), .84 (avoidance)). Test-rest reliability in the increased risk sample (A) was acceptable for the total score (r= .44, p= 0.011, n =33) and intrusion subscale (r= .61, p<0.001, n=35) but lower for the avoidance subscale (r=.40, p= 0.082, n=35).

Table 6.1- Mean scores on the Impact of Event scale in the increased risk and general population samples.

Measure	**Descriptive statistic**	**Increased risk sample (A)**	**General population sample (B&C)**
Impact of Event-Total score	n Mean (sd)	58 18.2 (12.69)	65 14.8 (15.15)
Impact of Event-Intrusion subscale	n Mean (sd)	60 8.1 (5.94)	68 5.9 (5.97)
Impact of Event-Avoidance subscale	n Mean (sd)	58 10.3 (8.47)	65 8.8 (9.68)

PART 2

EXPERIENCE QUESTIONNAIRE

6.3 THE DEVELOPMENT OF THE EXPERIENCE QUESTIONNAIRE

This questionnaire was developed prior to the researcher joining the psychosocial research group. Previous researchers in the group had derived initial items from issues raised in interviews with women attending the familial breast cancer clinic in Edinburgh and a limited review of the literature. A quantitative questionnaire had been developed and administered to women attending genetic counselling sessions. Following analysis of responses to these items a shortened version of the questionnaire had been devised. This questionnaire had been administered as part of the baseline assessment in a randomised controlled trial of two models of genetic counselling service provision. Data provided from this trial offered an opportunity for the current author to conduct a pilot study to assess the performance of the experience questionnaire and examine associations between experience and levels of general and cancer specific distress. This work is outlined as follows.

PILOT WORK

6.4 AIMS AND RATIONALE

Analysis was exploratory to determine if the experience measure could be utilised in further studies. The specific aims of this pilot work were:

- To determine if the questions were acceptable to women with a family history of breast cancer.
- To assess the distributional properties of the data obtained and ensure items were answered correctly.
- To explore associations between experience and levels of general and cancer specific distress.

Previous studies (reviewed in Chapter 2) have tended to assess associations between experience of breast cancer and either general or cancer specific distress. To clarify

the impact of experience on psychological adjustment in this population both general and cancer specific distress were examined and compared in this study.

Predictions were based on the literature reviewed in Chapter 2: General distress was expected to be more strongly associated with aspects of the experience having long term effects on the individual life and family. This included experience of bereavement from breast cancer in the family, multiple bereavement, maternal bereavement, age at bereavement and diagnosis and recency of these experiences (e.g. Dohrenwend and Dohrenwend 1974, Kurtz et al. 1997, Hopwood et al. 1998, Wellisch et al. 1992, Hopwood et al. 2001).

Cancer specific worry was predicted to be associated with experiences relating to personal risk, this included strong identification with a relative who had suffered breast cancer (Spira and Kenemore 2000). Positive role models of relatives who coped well with the disease and communication about breast cancer risk in the family was expected to be associated with reduced levels of cancer worry (Chalmers and Thompson 1996).

A number of aspects of experience were predicted to influence both levels of general distress and cancer worry. These included greater exposure to breast cancer, how traumatic and upsetting the experience had been overall as well as family tensions or changes caused by the illness experience (eg Lodder et al. 1999, Koocher 1986, McHorney and Mor 1988, Wellisch et al. 1992).

6.5 METHOD

6.5.1 Design

Data were collected in the context of a randomised controlled trial of two models of service provision in South East Scotland. Baseline data prior to genetic counselling are reported. A subset of data were released from the trial for this analysis.

6.5.2 Procedure

Participating General Practices in SE Scotland were randomised to receive the standard or novel service for patients whom they referred for genetic risk counselling. Referred patients were sent a postal questionnaire that they were required to complete and return before being allocated an appointment for risk counselling.

6.5.3 Participants

Women with a family history of breast cancer who had been referred for cancer risk counselling. Baseline data from a consecutive series of 96 women were available for use in this study.

6.5.4 Measures

6.5.4a Experience questionnaire

The questionnaire (please see Appendix II, page A-2) was organised in two parts:

Background information

The first two questions were aimed to assess the respondent's *personally relevant* family history of breast cancer rather than their *objective* family history. Participants were asked to report the number of close relatives they had personally known who had suffered from breast cancer and how many, if any of these relatives had died from breast cancer.

The participant was then asked to focus on one relative (their 'index relative') who had suffered from breast cancer and whose illness had particularly affected them (the participant).

The participant was asked a number of questions about their index relative:

- Their relation to them (coded categorically: 1= mother; 2= sister; 3= aunt; 4= gran; 5= cousin).
- How close they were (3 point Likert scale: 1= 'not at all close'; 2= 'quite close'; 3= 'very close').
- What age their relative was when first diagnosed with breast cancer (years).

- The participants age when their relative was first diagnosed (years).
- How the relative is now (coded categorically: 1= 'alive and well'; 2= 'alive but unwell'; 3= 'died from this cancer'; 4= 'died from other causes').
- If the relative had died, how long ago this happened (years).

Subjective experience items

Participants were then asked 11 questions about their subjective experience of their relative's illness. Participants were asked to provide answers regarding their lasting impression of this experience. These items were coded on a Likert scale: 1= 'not at all'; 2= 'a little'; 3= 'quite a lot'; 4= 'very much'.

- Did their illness/treatment cause them much physical suffering?
- Did their illness/treatment cause them much emotional distress?
- Overall, how upsetting was their illness for you?
- Overall, how well do you think they coped with their illness/treatment?
- Do you think you are like them in general?
- Do you feel that your relationship with this person changed when they developed cancer?
- How much do you feel your life plans have changed because of the risk of cancer in your family?
- Did you talk about your relative's cancer with the rest of your family at the time?
- Do you talk to friends now about your possible breast cancer risk?
- Do you talk now about your own risk with your family?

6.5.4b Distress

Measures of both general and cancer specific distress were included in this study.

General distress

The 30-item version of the General Health Questionnaire (GHQ-30) was used to assess general distress. This measure was described in Chapter 4 (section 4.4.4a, page 106). Responses were scored either a 0 or 1 (0, 0, 1, 1) and summed to achieve a total score. Using this method a score over 5 is taken as the cut-off point to indicate levels of distress that warrant further clinical assessment ('caseness') (Goldberg and Williams 1988).

Cancer specific distress

The cancer worry scale (described in Chapter 4 (section 4.4.4b, page 107)) was used to assess worry about breast cancer.

6.6 STATISTICAL METHODS

6.6.1 Descriptive statistics

Frequencies of responses for the experience items were examined to determine whether the questions had been correctly answered and to identify missing data that may indicate problematic items.

Descriptive statistics for scores obtained on the GHQ-30 and cancer worry scale by this sample are reported and GHQ 'cases' were identified. The internal consistency of the cancer worry scale was checked with Cronbach's alpha (Cronbach 1951) (see 6.1, page 128)

6.6.2 Associations between experience and levels of distress

In order to explore the data for associations between experience items and levels of distress a number of statistical techniques were employed depending on the nature of the items. For nominal items the chi-square test was used to determine differences in GHQ 'caseness' and t-tests were used to determine differences in cancer worry score.

The chi-square test compares the observed frequencies in each cell of a contingency table with the expected frequencies for each cell to test if the differences are due to chance. The chi-square statistic reflects the size of the difference between observed and expected frequencies, the greater the chi-square the more likely that the differences are significant. The chi-square test assumes that the observations in the contingency table are independent (ie each participant can have only one entry) and that the expected frequencies in each cell should be 5 or more. The chi-square test can only test for differences between the categories and is unable to test a one tailed hypothesis about direction of the effect.

The t-test aims to compare the amount of variability due to predicted differences in scores between the two groups compared to the total variability in scores (Green and D'Oliveria 1990). The difference in mean scores for both groups is compared to the overall variability in scores and statistic *t* represents the size of the difference between the means taking into account the total variance. The t-test is a parametric test and assumes that scores are measured on an interval scale, are normally distributed and variability of scores in each group is equivalent (homogeneity of variance). The t-test is fairly robust concerning these criteria and unless data divert substantially from the assumptions the results should remain valid (Green and D'Oliveria 1990).

T-tests and chi-square tests were also computed to determine differences in experience between individuals scoring above and below the median on the GHQ-30. For ordinal/interval items including the cancer worry score and subjective experience items Pearsons correlation coefficient was computed to determine the association between distress and experience. Correlation coefficients vary between –1.0 and +1.0. Both extremes represent perfect relationships between variables whereas as coefficient of 0.0 represents no relationship. A positive coefficient means that a higher score on one variable is associated with higher scores on the second variable. A negative relationship means that high scores on one variable are associated with low scores on the other variable. Intercorrelations were examined using Pearsons product-moment correlation coefficient. This is a correlational technique suited for use on interval scales. The variables need not be normally distributed but the technique does assume linearity of relationship.

A probability value of $p<0.05$ was taken as significant. Trends of $p<0.1$ are also reported given the exploratory nature of this analysis.

6.7 RESULTS

6.7.1 Sample

The sample consisted of 96 women with a mean age of 38 years (sd= 10.41, range = 19 - 68 years). The majority of women were married or living with a partner (68.6%)

and 21.9% had never been married. Just fewer than twenty percent (19.8%) were university graduates with over half of the sample (51.1%) having received further education or training beyond the age of 18.

6.7.2 Descriptive statistics

6.7.2a Experience questionnaire

Background items

The vast majority of women reported to have personally known at least one close relative who had suffered breast cancer (97%). Forty-six women (49 %) had known >1 relative who had suffered from breast cancer (range= 0-6). Fifty-six women (60%) reported that at least one of these relatives had died from breast cancer (range 0-4).

The majority of index relatives were mothers (49%), sisters (21%) or aunts (18%). Sixty-eight percent of women reported to be very close to their index relative and 23% quite close. The mean age of the index relative at diagnosis was 45.1 years (range= 25-70, sd= 9.92), and the mean age of the participant when the index relative was diagnosed was 23.1 years (range < 1- 62, sd= 12.04). Fourteen women (15%) were under age 11 when their index relative was diagnosed, 22 (23%) were aged between 11-20 and 45 (47%) were 21 or older. Selecting those women who chose their mother as the index relative, the proportion of women in each age category was 24% (0-10), 43% (11-20), 33% (21+). Fifty percent of women reported that their index relative had died from cancer and 4% that their index relative had died of other causes. The mean time since death was 15.1 years (range = < 1-40 years, sd= 12.01). Twenty-eight percent reported that their relative was alive and well and 15% reported that their relative was alive but unwell.

Subjective experience items

Table 6.2 shows the frequencies of the subjective experience items. A number of items appeared either negatively skewed (1, 2, 3, 4) or positively skewed (6, 7, 8).

Table 6.2- Frequencies of subjective experience items

Item	n %	Not at all	A little	Quite a lot	Very much	Missing
1. Did their illness/treatment cause them much physical suffering?		-	11 11.5	43 44.8	32 33.3	10 10.4
2. Did their illness/treatment cause them much emotional distress?		-	10 10.4	30 31.3	46 47.9	10 10.4
3. Overall, how upsetting was their illness for you?		3 3.1	12 12.5	16 16.7	55 57.3	10 10.4
4. Overall, how well do you think they coped with their illness/treatment?		3 3.1	4 4.2	47 49	33 34.4	9 9.4
5. Do you think you are like them in general?		11 11.5	28 29.2	23 24	23 24	11 11.5
6. Do you feel that your relationship with this person changed when they developed cancer?		44 45.8	21 21.9	18 18.8	5 5.2	8 8.3
7. Do you feel that your role in the family changed because of their cancer?		39 40.6	25 26	11 11.5	12 12.5	9 9.4
8. How much do you feel your life plans have changed because of the risk of cancer in your family?		41 42.7	30 31.3	10 10.4	7 7.3	8 8.3
9. Did you talk about your relative's cancer with the rest of your family at the time?		22 22.9	33 34.4	23 24	12 12.5	6 6.3
10. Do you talk to friends now about your possible breast cancer risk?		35 36.5	42 43.8	9 9.4	3 3.1	7 7.3
11. Do you talk now about your own risk with your family?		31 32.3	46 47.9	11 11.5	5 2.1	6 6.3

6.7.2b Distress

General distress

The mean score in this sample was 4.9 (sd= 7.19) and the median score was 1. Twenty-eight women (29.2%) scored above the cut off point of 5 on the GHQ-30, indicating possible clinical levels of distress.

Cancer worry

Ninety-three women fully completed the cancer worry scale. The mean score on the cancer worry scale was 11.2 (sd= 2.81, range = 6 – 18). The data showed a normal distribution and no outliers were found (skew = .34, kurtosis = -.23). The Cronbach's alpha value was .83 suggesting that the scale showed good internal reliability.

6.7.3 Distress and background demographics

General distress and cancer worry were not significantly correlated with age ($p<0.05$) and were not associated with marital status or education ($p<0.05$).

6.7.4 Experience and distress

6.7.4a General distress

There were no significant differences between GHQ 'cases' and 'non cases' on any experience items. Two trends were found. 'Cases' reported that their index relative was younger at diagnosis ($p= 0.07$) and felt their role in the family had changed because of their cancer ($p= 0.07$).

For the sub-sample of women who chose their mother as the index relative (n= 42) GHQ 'cases' were older when their mother was diagnosed (mean age= 21.5 years, sd= 9.86) than 'non cases' (mean age= 15.0 years, sd= 8.03) ($t= 2.33$, $df= 40$, $p= 0.03$). When looking at levels of distress by summing scores (0, 0, 1, 1) there was a significant correlation between GHQ score and age at mother's diagnosis ($r= .38$, $p=0.01$, n= 42). There was no difference between the three age categories (<11, 11-20, > 20) on GHQ 'caseness' or levels of distress for the whole sample or the sub-sample who chose their mother as the index relative ($p<0.05$).

Table 6.3 indicates a number of differences in experience items between individuals who scored above and below the median on the GHQ-30. Participants who scored ≥ 2 were significantly more likely to report that they felt their relationship with the index relative and role in the family changed because of the index relatives cancer and to have found their illness more upsetting ($p<0.05$). Participants whose index relative had died were more distressed if this had occurred more recently ($p<0.05$).

Table 6.3- Differences in experience between women scoring above and below the median on the GHQ-30.

Experience item	t	df	p		GHQ score = 0 or 1	GHQ score ≥ 2
Do you feel that your relationship with this person changed when they developed cancer?	3.04	86	0.003	n Mean sd	44 1.5 0.79	44 2.1 1.01
Do you feel that your role in the family has changed because of their cancer?	2.69	85	0.008	n Mean sd	45 1.7 0.98	42 2.3 1.08
Recency of bereavement of index relative (if any) (years)?	2.60	49	0.012	n Mean sd	26 19.2 11.52	25 10.9 11.21
Overall, how upsetting was their illness for you?	2.33	84	0.022	n Mean sd	42 3.2 0.98	44 3.6 0.68

6.7.4b Cancer worry

There were no significant differences in cancer worry scores between participants with different responses to the categorical experience variables. A number of the subjective experience items were found to be associated with cancer worry. Table 6.4 shows the Pearson correlation coefficients between the cancer worry scores and experience items. Cancer worry scores were significantly positively associated with the extent to which participants felt their life plans had changed because of the risk of cancer in their family and the degree of which their role in the family had changed ($p<0.05$). Cancer worry was also significantly positively correlated with the degree to which the participant talks about their risk with their family ($p<0.05$). Trends also suggested cancer worry was higher for participants who talk to their friends about their risk of breast cancer and for women who found their relatives illness more upsetting ($p<0.1$).

Table 6.4- Correlations between cancer worry and experience items

Experience item	n	r	p
How much do you feel your life plans have changed because of the risk of cancer in your family	88	.48	0.000
Do you talk now about your own risk with your family?	90	.23	0.033
Do you feel that your role in the family changed because of their cancer?	87	.23	0.034
Do you talk to friends now about your possible breast cancer risk?	89	.20	0.067
Overall, how upsetting was their illness for you?	86	.19	0.08

6.8 SUMMARY AND DISCUSSION

6.8.1 Acceptability of the experience questionnaire

This pilot work provided useful information regarding the performance of the experience questionnaire. Item frequencies did not reveal major problems with missing data and suggested that the items were acceptable to the majority of women in this sample. The results did however suggest some changes to the coding and scaling of items that may improve the questionnaire. The item assessing the closeness of relationship between the participant and index relative was skewed with 68% of participants reporting to be 'very close' to their index relative. In order to improve the spread of responses an additional category 'extremely close' could be used in subsequent research. The question assessing recency of bereavement of the index relative was coded in years and ranged from less than 1 year to 40 years ago. It may be more sensitive to code this measure in months rather than years. The 11 Likert items referring to women's subjective experience of their relatives illness were also skewed to end points and a 5 point scale ranging from 'not at all' (1) to 'extremely' (5) may improve the distribution of the data.

6.8.2 Experience and distress

In the full sample GHQ 'caseness' was not significantly associated with any experience items although the *level* of general distress was. A median split revealed that levels of general distress were associated with some of the experience items in the expected direction. This suggests that the Likert scoring method on the GHQ-30

(1, 2, 3, 4) may be more sensitive in detecting associations with experience items than scoring method used in this pilot work (0, 0, 1, 1) (See section 4.4.4a, page 106).

Higher levels of general distress were associated with greater changes in the participant's relationship with their index relative and changes in family roles because of breast cancer in the family. This confirms the findings of Wellisch et al. (1992) whose qualitative study indicated that adjustment problems in women at increased risk of breast cancer were associated with the changes in life plans created by breast cancer in the family.

Wellisch et al. (1992) also observed women who were younger at the time of their mothers diagnosis, particularly adolescents showed great problems in adjusting to their mothers breast cancer, although other studies have reported no effect of age at maternal diagnosis on levels of cancer specific distress (Erblich et al. 2000). In this study no association was found between participants' age when their index relative was diagnosed and level of general or cancer specific distress. However this study was not examining the effect of maternal breast cancer alone but instead asked participants to refer to the experience of a relative that had particularly affected them. Approximately half of the participants chose a relative other than their mother. The age at which other breast cancer experiences occur may not have such a dramatic effect on women as maternal breast cancer at adolescence. Among participants who had chosen their mother as their index relative, a *positive* association was found between age at diagnosis and levels of general distress, suggesting that those who were older when they lost their mother have higher levels of distress. This may represent the recency of this experience. Women who were older when their mother was diagnosed are likely to have experienced this event more recently. Alternatively it is possible that women who were children or adolescents at the time of a breast cancer event did not understand what was happening or were deliberately protected from the events by other family members. No difference between adolescents and other age groups were observed. Differences in the distribution of ages may help explain the inconsistencies of the findings. In this study a small proportion of women were adults (21+) at the time of mothers diagnosis (n=14) whereas Wellisch et al.

(1992) reported 60% of their sample to be in this age category. In order to clarify inconsistencies of the literature in this area it is important to examine the role of factors that may moderate the impact of age including risk status and awareness or understanding of breast cancer events at the time. When looking at the age of the index relative at diagnosis a trend was found to suggest that the index relatives of GHQ 'cases' were diagnosed at a younger age than those of 'non cases'. This effect has also been shown in previous studies (Erblich et al. 2000, Lodder et al. 1999). However since age at diagnosis is one of the risk criteria it is unclear if this effect represents differences in experience of the disease or risk status.

It was surprising that general distress was not associated with other aspects of bereavement (including experience of bereavement, multiple bereavement and maternal bereavement). Loss of an index relative from breast cancer was not associated with general distress or cancer worry. Associations have been reported between cancer bereavement and intrusive thoughts about cancer in women at increased risk of breast cancer (Zakowski et al. 1997, Erblich et al. 2000) although no effect has been reported on levels of cancer worry (Hopwood et al. (2001). Bereavement may therefore influence intrusive thoughts about cancer rather than general distress or worry over personal risk. Leedham and Meyerowitz (1999) also suggest that past bereavement due to cancer may invoke subtle existential concerns in adulthood rather than general distress assessed by global distress measures.

As predicted general distress was higher for women who had lost their index relative to breast cancer more recently and those who found the illness more upsetting. Previous qualitative studies have indicated that the more emotional and intense the breast cancer experience the harder it can be to resolve (Chalmers and Thompson 1996) and that emotional descriptions of the impact of the disease are associated with increased distress (DudokdeWit et al. 1997). It is unlikely that the association between recency of bereavement and distress represents a *normal* bereavement response. The average time since bereavement of the index relative in this sample was 15 years. Persistent grief over 1 year has been considered an abnormal response (Koocher 1986). It is more likely that this effect may instead reflects a *reactivation*

of grief shown in previous studies of women attending for genetic counselling for breast cancer (DudokdeWit et al. 1997, Lodder et al. 1999, Hopwood et al. 1998).

6.8.3 Cancer worry scale

The study revealed that the cancer worry scale was acceptable to this sample. The scale showed good internal reliability and scores were associated with aspects of experience of breast cancer in the family. As predicted cancer worry was positively associated with changes in life plans and family roles because of breast cancer in the family. Cancer worry was also associated with communication about cancer risk within the family but in the opposite direction to that predicted. Communication about risk was predicted to be associated with lower levels of cancer worry but instead a positive association was found. Hilton (1996) found that for some families, talking and sharing concerns about breast cancer helped them return to normal, whereas for others communication about breast cancer was limited and did not reduce anxiety. In this study it is possible that increased talk about cancer risk reflected greater cancer worry or that communication about breast cancer itself increased fear and anxiety about the disease (Chalmers et al. 1996). A cross sectional study is unable to determine causality between these variables and further prospective study of communication frequency and style is required to address this issue further.

A number of items did not show predicted association with cancer worry. Cancer worry was not associated with identification with the index relative or perceptions of how well the relative had coped with the disease. It is possible that single items are not able to fully tap the complex issues surrounding identification with an affected relative or their ability to cope with breast cancer. Exposure to breast cancer represented by the number of affected relatives personally known was not associated with either general or cancer specific distress as has been reported in previous studies (Wardle 1995, Baider et al. 1999). In addition, perception of physical suffering and emotional distress experienced by the index relative was not associated with measures of distress. This may be due to the skewed distribution of these items or that they do not adequately assess exposure to the relative's illness.

6.8.4 Methodological issues

A further methodological issue in this study was the fact that the sample had not received genetic counselling when they completed the questionnaire. Women in this situation may have been more concerned about attending the familial breast cancer clinic and learning about their risk status at this time, making effects of experience of levels of distress difficult to determine.

A number of elements of experience were omitted from the questionnaire that may have proved important in this sample. For example the effect of caring for relatives (Erblich et al. 2000), exposure to late stages of the disease (Lodder et al. 1999), if the index relative was currently undergoing treatment, confidence in medical care for breast cancer and experience of other family members (Smith et al. 1999). Positive aspects of the experience and positive breast cancer outcomes that may act as buffers were also not included. The experiences of women with a family history of breast cancer are likely to be diverse and a questionnaire is unable to cover all possible experiences. Given the constraints on questionnaire length the current questionnaire attempted to measure aspects of experience which had been indicated as important in the literature and previous interviews with women at increased risk in Scotland.

In conclusion, the format of the experience questionnaire worked well and items were answered correctly in this study. A number of aspects of experience were found to show predicted associations with levels of general and cancer specific distress in this sample. Improvements could be made to some of the response formats and additional questions included. It was decided that following further refinement this measure could be utilised in the main study.

6.9 ADAPTATION OF THE EXPERIENCE QUESTIONNAIRE

A number of modifications to the experience questionnaire were highlighted in the pilot work. This section discusses and justifies changes made to the experience questionnaire and outlines how the measure performed in the main study. Descriptive statistical results are reported and where appropriate comments are made regarding comparison of results to those obtained in the previous pilot work. A copy of the adapted questionnaire is included in Appendix II (page A-4)

The pilot work showed that the experience questionnaire was acceptable to women with a family history of breast cancer and showed predicted associations with levels of general and cancer specific distress in women with a family history of the disease. However, the results suggested a number of potential improvements. These included coding recency of bereavement in months rather than years to improve the sensitivity of the measure. The response scale for the subjective experience items was also expanded from a 4 point Likert scale item to a 5 point scale. Streiner and Norman (1991) provide data that indicates Likert scales of less than 5 points result in loss of information and show poor reliability. Expanding the scale should therefore enhance the reliability of the items as well as improving the distribution of data.

The primary aim of the research reported in this thesis was to test the mediation model that perceptions of breast cancer mediate the impact of experiences of the disease in the family on psychological well-being in women at increased risk (Fig 4.1, page 100). The experience items therefore needed to focus on aspects of experience likely to influence women's perceptions of breast cancer.

The large number of variables included in the questionnaire was problematic because of the potential number of statistical analyses to be conducted. Multiple testing may lead to significant effects occurring by chance. In addition, high intercorrelations between experience items may lead to problems of multicollinearity in future multiple regression analysis (see Table 4.1, page 102). In order to counteract these problems it was decided to reduce the number of subjective experience items using item-scaling analysis. Factor analysis was originally considered as a technique used for item reduction (Tabachnick and Fidell 1996). Factor analysis was performed on

the data but a cohesive factor structure could not be identified. A number of limitations regarding the application of factor analysis to the data set were thought to account for this result. Firstly, the measure was not designed as a psychometric assessment of experience but rather a pragmatic measure of specific issues previously identified. It was not assumed therefore that the items would fully capture underlying latent variables. Secondly, the variables in the measure are likely to be complexly interrelated. In factor analysis pure variables correlated with only one factor are preferred. Tabachnick and Fidell (1996) warn that measures that include variables of differing complexity are likely to form factors that do not reflect underlying processes. Thirdly, the sample size was low in regards to the general rule of thumb for factor analysis (n>300) and borderline in terms of the expected ratio of cases to items (Ferguson and Cox 1993, Tabachnick and Fidell 1996). It was therefore decided to group the items in terms of face validity and conduct item-scaling analysis to ensure the items were compatible

6.9.1 Additional background items

The literature has indicated the importance of experiences of bereavement and diagnosis in the family (eg Kelly et al. 1987, Hilton 1993, Zakowski et al. 1997, Erblich et al. 2000). Recent experiences are likely to have a large impact on availability of breast cancer images and perceptions of the disease. A recent diagnosis of breast cancer may have a strong effect on levels of distress and representations of the disease. It was therefore decided to assess not only the recency of bereavement of the index relative from breast cancer but also the recency of bereavement of any of the relatives personally known to the respondent. Questions concerning recency of diagnosis of breast cancer in the family were also included.

6.9.2 Adaptation of subjective experience items

The response frame for these items was expanded from 1 (not at all) – 4 (very much) to 1 (not at all) – 5 (extremely). Full versions of the original questionnaire used in pilot work and adapted questionnaire used in the main study are provided in the Appendices. Although the first 4 items (*'Did their illness/treatment cause them much physical suffering?', 'Did their illness treatment cause them much emotional distress?', 'Overall how upsetting was their illness for you?', 'Overall how well do you think they coped with their illness/treatment?'*) were not strongly associated with

distress in the previous study, the sample was assessed prior to genetic counselling and it is possible that the items may be associated with distress after women are informed of their risk status. These items were also considered potential determinants of women's perceptions of breast cancer. It was therefore decided to retain these items in the questionnaire.

From a theoretical perspective perceived resemblance with an affected relative may be a strong predictor of response to genetic risk (see section 4.2, page 73). However item 5 *('Do you think you are like them in general?')* was not associated with distress in the previous study. This may be because a single general item is not adequate to assess perceived resemblance. Additional more specific items may create a more reliable measure of resemblance. Two questions were therefore derived from components of resemblance highlighted as important in the literature (Davison 1989, Richards and Ponder 1996, Richards 1996): *('Do you think you are like them in body shape and size?' and 'Do you think you are like them in personality?').*

Items 6-8 showed strong associations with distress and were also predicted to be associated with illness perceptions. These items were therefore kept in the questionnaire.*('Do you feel that your relationship with this person changed when they developed cancer?', 'Do you feel that your role in the family had changed because of their cancer?', 'How much do you feel your life plans have changed because of the risk of cancer in your family?'*) However item 6 referring to 'change' in relationship with the index relative was ambiguous and may refer to positive or negative changes. Hilton (1996) found that some families with a patient suffering from breast cancer talked about developing closer bonds because of the experience whereas others described the breaking apart of relationships. The item was intended to refer to detrimental effects of the experience on the relationship following work by Wellisch et al. (1996). Since this item was positively associated with levels of general distress in the previous study it appears to have been interpreted in that manner. In order to clarify this item in subsequent research the item was reworded to *' 'Do you feel that your relationship with this person* ***deteriorated*** *when they developed cancer?'.*

The final 3 items were omitted from the questionnaire. (*'Did you talk about your relatives cancer with the rest of your family at the time?', 'Do you talk to friends now about your possible breast cancer risk?', 'Do you talk now about your own risk with your family?'*). These items had been predicted to show negative associations with distress based on previous research indicating that open communication about cancer was associated with better adjustment of family members (Leedham and Meyerwitz 1999, Chalmer and Thompson 1996, Chalmers et al. 1996). However two of these items were found to show moderate positive associations with cancer worry. This conflicting result highlights the diverse nature and effects of communication in these circumstances. It is difficult to assess the impact of communication about cancer or cancer risk on illness perceptions or levels of distress without an understanding of the content and type of communication (ie open or restricted). Family communication about genetic risk therefore needs to be examined in more detail than is possible in the current study.

It was decided to include positive aspects of experience in order to determine if positive experiences have a beneficial effect on illness perceptions and/or levels of distress. Theoretically, it is plausible that positive experiences may promote more positive representations of the disease in question. Studies in the literature have shown individuals to report positive effects of cancer in the family. Petrie et al. (1999) found breast cancer patients reported a number of positive aspects of their illness experience including improved quality of close relationships, greater appreciation of life, change in personal priorities and greater empathy for others. Taylor and Armor (1996) discuss the use of positive illusions as a strategy for coping with threatening events. They review research that suggests patients suffering from a range of illnesses (including cancer) often react to threatening situations by perceiving positive changes (eg gaining personal qualities, increased understanding of others, finding meaningfulness in life etc). This provides a protective mechanism by which to bolster ones self perception in the face of adversity. They propose that a positive assessment of the event, perception of control and sense of optimism about the future might promote adjustment. Similar strategies may be seen in relatives of patients. Caregivers of terminal cancer patients have reported positive feelings concerning the time and interaction with the affected relative (Grbich et al. 2001).

Leedham and Meyerowitz (1999) found 93% of adults who reported retrospectively on their experiences of parental cancer reported at least one positive change in their lives because of the experience. Two thirds reported improvements in their relationship with the sick parent and a third with the healthy parent. Finally, Savage and Clarke (1998) found older women often cited positive examples of breast cancer patients observed within their social circle when asked for beliefs regarding the treatment and cure of breast cancer. This suggests that observing positive experience of breast cancer may also influence healthy women's perceptions of the disease. In order to examine the impact of positive experiences of women at increased risk of breast cancer three further items reflecting positive experiences that had been prominent in the interview data and were consistent with previous research findings were added to the questionnaire. *('Did this person hold a positive attitude towards their illness?', 'Do you feel that your experiences have brought the family closer together?', 'To what extent have your experiences been positive?')*

6.10 DESIGN AND SAMPLE

Data for this analysis were derived from the cross-sectional study outlined in Chapters 4 and 5. Results from the increased risk sample (Sample A) are reported. (Please see Chapter 5 (5.2.1, page 116) for a description of the sample).

6.11 STATISTICAL ANALYSIS

6.11.1 Descriptive statistics

Response distributions and descriptive statistics were examined in order to help interpret if the items appeared to work and to check whether responses to items were normally distributed and hence whether parametric statistics could appropriately be used throughout the analysis presented in this thesis.

6.11.2 Item grouping

Items were grouped in terms of face validity and item-scaling analysis conducted to ensure the items were compatible. Highly correlated items could then be summed for use in future analysis. This technique has been recommended as a logical way to reduce problems of multicollinearity in multiple regression analysis (Everitt 1996).

From the 12 subjective items used to assess experience in the increased risk sample five subscales were predicted: 'traumatic experience', 'coping', 'resemblance', 'change' and 'positive experience'. These subscales and constituent items are described in Table 6.5

In order to assess the internal consistency of the item grouping, item convergent and discriminant validity were examined. This technique is widely used in test construction and for checking the homogeneity of items in scales (Kline 1986, Streinder and Norman 1991). Kline (1998) describes this technique as a highly efficient, useful procedure when data are not suitable for factor analysis. It has been used for developing clinical measures including quality of life in cancer patients (Cull et al. 2001). Three statistics are examined, internal consistency, item convergence and item discriminance. Internal consistency is examined using Cronbach's alpha. Item convergence tests the proportion of items that correlate highly with the scale (when the item in question is omitted). Item discriminant validity compares the strength of correlations between items with their own scale and other scales (Nunnally and Bernstein 1994). These tests are defined below.

- **Item convergent test** = the number of item-scale correlations (with the item omitted) >0.4/total no of correlations.
- **Item discriminant validity test** = number of times in which an item correlates more highly with its own scale (with the item omitted) than with another scale/total number of correlations.

Table 6.5- Grouping of items from the experience questionnaire.

Predicted Subscale	Description of subscale	Subjective experience item	
Traumatic experience	Extent to which the experience was upsetting for the index relative and the respondent.	1	Did their illness/treatment cause them much physical suffering?
		2	Did their illness/treatment cause them much emotional distress?
		3	Overall, how upsetting was their illness for you?
Coping	Extent to which the index relative coped positively with breast cancer.	4	Overall how well do you think they coped with their illness/treatment?
		5	Did this person hold a positive attitude towards their illness?
Resemblance	Extent to which the respondent feels they resemble the index relative physically and in personality.	6	Do you think you are like them in body size and shape?
		7	Do you think you are like them in personality?
Change	Extent to which the respondents family life and plans have changed because of their experiences.	8	Do you feel that your relationship with this person deteriorated when they developed cancer?
		9	Do you feel that your role in the family has changed because of your experiences of breast cancer?
		10	How much do you feel your life plans have changed because of the risk of cancer in your family?
Positive experience	Positive aspects created by the experience.	11	Do you feel that your experiences have brought the family closer together?
		12	To what extent have your experiences been positive?

6.12 RESULTS

6.12.1 Descriptive statistics

This analysis refers to the 117 participants in the increased risk sample (Sample A) (see Chapter 5, 5.2.1, page 116).

6.12.1a Background items

Few questions had missing data. Two participants omitted the question '*Have any of these relatives been diagnosed with breast cancer recently?*' One participant omitted the question '*How many, if any of these relatives have died because of breast cancer?*' Three participants omitted the question '*Please could you let us know how long ago these relatives died?*' Two participants omitted the question '*What age were they when breast cancer was first diagnosed?*'

On average participants personally knew 2 relatives who had suffered from breast cancer (mean =2.1, sd =1.25, range =1-7). Thirty-eight (32.5%) reported that a relative had been diagnosed within the past 5 years. Eighty-eight (75.2%) reported that they had at least one relative who had died from breast cancer (mean =1.1, sd = 1.02, range =0-6). The average time since the most recent bereavement was 16.3 years (sd =11.93, range =1-49 years).

The majority of participants referred to their mother as the index relative on the personal experience questions (69.2%). Other participants referred to sister (18.8%), aunt (8.5%), cousin (2.6%) and grandmother (0.9%) as the index relative. The average age at which the index relative had been diagnosed with breast cancer was 44.0 years (sd =10.3, range 0-47). The average age of participants when their relative was diagnosed was 21.7 years (sd =11.74, range =0-47).

Seventy-one (60.7%) participants reported that their relative had died from breast cancer and 6.0% had died from other causes. Thirty-six (30.8%) women reported that their relative was alive and well and 2.6% alive but unwell. For women who had lost their relative from breast cancer the average time since bereavement was 15.6 years (sd =11.0, range =3months – 38 years).

The results obtained from the background experience items in this study showed some similarities to those reported in the pilot work. For example, the mean age of the index relative at diagnosis (mid forties) and age of the participant (early twenties) was comparable and the average time since bereavement of the index relative in both samples was approximately 15 years. Slightly more women in this sample however reported to know at least 2 relatives who had been affected by breast cancer (59%) compared to the previous study (49%). Also, slightly more women had lost at least one relative to breast cancer (75%) than in the previous study (60%) and a larger proportion of women in this sample reported that their index relative had died from breast cancer (60%) than in the previous study (50%). Women in this sample were also more likely to have chosen their mother as the index relative (69%) compared to the previous sample (49%). These differences most probably reflect the differences in risk status between the samples. Women in this study had received genetic counselling and were at significant increased risk of breast cancer whereas those in the previous study were attending a community based genetic service in order to determine their risk status. A proportion of the previous sample may not have had a significant family history.

6.12.1b Subjective experience items

Table 6.6 shows the descriptive statistics of the subjective experience items. Extending the response format for these items improved the spread of the data in comparison to the pilot work reported in Chapter 6 (Part 2, sections 6.3-6.8). In both studies few participants responded in the categories 'not at all' or 'a little' for the first 4 items. The results reported in Table 6.6 show that responses were more evenly spread over the 3 remaining categories rather than being confined to 2 categories ('quite a lot' and 'very much') in the previous version of the questionnaire. The resemblance items (6&7) also appeared to benefit from a 5 point scale. In the previous study 24% of respondents reported themselves to be 'very much' like their index relative in general. In the present study 10% reported they thought they were 'extremely' like their index relative physically and in personality. Item 8 concerning relationship with the index relative showed a large change in distribution of responses. In the previous study 45.8% reported that their relationship with their

index relative had 'not at all' 'changed' whereas in the present study 80.3% reported that their relationship with their index relative had 'not at all' 'deteriorated'.

Table 6.6- Distributional properties of subjective experience items utilising a 5-point scale

Subjective experience item.	Missing data	Mean	sd	Distribution of responses Number of respondents Percent				
				Not at all	A little	Quite a lot	Very much	Extremely
1. Did their illness/treatment cause them much physical suffering?	3	3.8	1.01	1 0.9	13 11.1	30 25.6	39 35.3	31 26.5
2. Did their illness/treatment cause them much emotional distress?	6	3.9	0.91	-	6 5.1	36 30.8	36 30.8	33 28.2
3. Overall, how upsetting was their illness for you?	3	4.2	0.98	1 0.9	6 5.1	21 17.9	25 21.4	61 52.1
4. Overall how well do you think they coped with their illness/treatment?	4	3.8	1.05	4 3.4	8 6.8	26 22.2	42 35.9	33 28.2
5. Did this person hold a positive attitude towards their illness?	6	3.6	1.16	7 6.0	12 10.3	27 23.1	37 31.6	28 23.9
6. Do you think you are like them in body size and shape?	3	2.8	1.31	23 19.7	32 27.4	19 16.2	28 23.9	12 10.3
7. Do you think you are like them in personality?	3	2.8	1.20	17 14.5	34 29.1	33 28.2	18 15.4	12 10.3
8. Do you feel that your relationship with this person deteriorated when the developed cancer?	3	1.3	0.84	94 80.3	9 7.7	6 5.1	3 2.6	2 1.7
9. Do you feel that your role in the family has changed because of your experiences of breast cancer?	2	2.4	1.40	42 35.9	27 23.1	13 11.1	22 18.8	11 9.4
10. How much do you feel your life plans have changed because of the risk of cancer in your family?	2	1.8	1.06	59 50.4	31 26.5	14 12.0	8 6.8	3 2.6
11. Do you feel that your experiences have brought the family closer together?	2	2.6	1.27	25 21.4	38 32.5	23 19.7	17 14.5	12 10.3
12. To what extent have your experiences been positive?	2	2.5	1.13	26 22.2	37 31.6	30 25.6	17 14.5	5 4.3

6.12.2 Item scaling analysis

A summary of the analysis is provided in Table 6.7. (Please see Appendix III, page A-20, for full reports of item-scale correlations). Cronbach's alpha values were reasonable for the first 3 scales ('traumatic experience', 'coping' and 'resemblance') but very low for the last 2 scales ('change' and 'positive experience'). The item convergent tests for the 'traumatic experience ', 'coping' and 'resemblance' subscales were good with all items correlating r >.4 with their subscale. The 'change' and 'positive experience' subscales failed the item convergent test with none of the items correlating r >.4 with their subscale. The 'change' subscale did also not perform well on the item discriminant test. (There were 3 cases in which an item on this subscale correlated more strongly with another subscale than the 'change' subscale). These results suggest that the 'traumatic experience', 'coping' and 'resemblance' subscales appear reliable, whereas the 'change' and 'positive experience' subscales do not.

The change scale was assessed with each item in turn deleted. In particular the scale was examined with item 8 omitted. This item was non-discriminatory and may have caused problems in the scale. However even with this item omitted the alpha for the scale was still low (.35) and correlation between the remaining items still not adequate (r= .22, p= 0.018). It was therefore decided that the items from the 'change' and 'positive experience' subscales should remain as single items.

Table 6.7- Internal reliability, item convergent and discriminant tests for proposed subscales

Proposed Subscale Scale	No. of items	Alpha	Corrected item-scale correlations	Item convergent test	Item-scale correlations (range excluding own scale)	Item discriminant test
Traumatic experience	3	.70	.42, .62, .63	3/3	-.01 to .51	11/12
Coping	2	.72	.57	2/2	-.01 to .36	8/8
Resemblance	2	.69	.51	2/2	.16 to .40	8/8
Change	3	.36	.18, .23, .30	0/3	.02 to .39	9/12
Positive experience	2	.35	.40	0/2	.086 to .40	8/8

6.13 SUMMARY AND DISCUSSION

The experience questionnaire was adapted in light of the results from the pilot work (Chapter 6, Part 2, sections 6.3-6.8) and theoretical perspectives (Chapter 3,) in order to address the aims and hypotheses of this thesis (Chapter 4, section 4.1). A major change was increasing the scale of the subjective experience from a 4 point to a 5 point scale in order to improve the discriminatory power of the items and distribution of data. Examination of the distribution of comparable items in both studies revealed that the spread of the data was improved. However, item 7 (*Do you feel that your relationship with this person deteriorated when they developed cancer?*) showed a stronger positive skew than the comparable item in the previous study referring to relationship 'change'. Although the question was reworded in order to reflect negative changes in the relationship the results suggested that the word 'deteriorated' may have been too harsh to reflect more subtle changes in the relationship. The new items assessing positive experiences in this study (items 11&12) appeared to be answered correctly and showed good distribution across response categories.

The subjective experience items were reduced into groups for pragmatic statistical reasons. Three of the five proposed scales were deemed acceptable following scaling analysis and will be used in subsequent research. The remaining items will be analysed as single items. There are a number of possible reasons why two of the scales failed the analysis. Firstly, the proposed construct may not exist; secondly, the items may have been inadequate and thirdly, the construct may be factorially complex (Kline 1998). In this study the scales were developed based only on face validity. It is possible that the predicted dimensions do not represent true dimensions of experience. For example, items proposed to reflect the 'change' subscale included both past changes in family roles and relationships created by breast cancer and future changes to life plans following breast cancer risk. These aspects are likely to represent different issues and require separate assessment. The 'positive experience' subscale only comprised of two items. It is likely that this was insufficient to cover all positive aspects of the experience. Although item scaling was useful in this context the nature of the technique has been criticized due to its circularity (Kline 1998). Whilst the analysis can indicate that items assess a similar construct it provides no information regarding the validity of that construct. This analysis

provided acceptable sub-scales representing certain aspects of experience including trauma, coping and issues resemblance relevant to identification. These item groupings cannot be regarded as true 'scales'. There is no evidence that item groupings derived represent all aspects of the dimension or are valid measures.

In order to develop psychometric measures of dimensions of experience of breast cancer more detailed and thorough work needs to be conducted. Qualitative research should initially be conducted to explore specific aspects of experience in depth and derive a wide range of possible items. These items can then be used to develop a scale of the dimension of experience in question and ensure than the scale includes all variables that represent the dimension. Item analysis or factor analysis can then be conducted to explore the hypothesised scale structure and determine appropriate items that collectively assess the specific aspect of experience in question. Finally, the reliability of the scale should be assessed and tests of its validity conducted.

PART 3

ADAPTING THE IPQ-R FOR USE IN HEALTHY SAMPLES

6.14 THE REVISED ILLNESS PERCEPTION QUESTIONNAIRE (IPQ-R): BACKGROUND

The revised version of the IPQ (IPQ-R) was described in Chapter 4 (4.4.4). This is a theoretically driven measure designed to assess illness perceptions across a range of illnesses (Weinman et al. 1996, Moss-Morris et al. 2002). The measure assesses the dimensions outlined in earlier work on illness representations (identity, consequences, timeline, control/cure, cause) as well as a number of new components (timeline cyclical, illness coherence, emotional representations). For a description of dimensions please see Figure 6.1.

The identity dimension is assessed by a list of 14 symptoms. Respondents are asked to rate whether or not they believe each of these symptoms to be specifically related to their illness. The number of symptoms associated with the illness is summed to

obtain an identity score. Causal items are assessed using a checklist of 18 potential causes of illness. The respondent is asked to rate whether or not they believe each of these factors were causes of their illness on a 5 point Likert scale. Respondents are also asked to report what they believe to be the three most important causes of their illness.

Seven dimensions of illness representations (timeline acute/chronic, timeline cyclical, consequences, personal control, treatment control, illness coherence and emotional representations) are assessed using Likert items rated on a 5 point scale ranging from strongly agree to strongly disagree. Items for these subscales are summed (after reverse scoring as necessary) and the mean value calculated. Where items are omitted no subscale score is derived.

Originally these 7 dimensions of the IPQ-R were assessed using 50 items. This questionnaire was available from the author and showed promising psychometric properties (Weinman, personal communication, 1999). Validation studies of the IPQ-R were conducted subsequent to the design and data collection for this thesis. The validation work on the IPQ-R supported the factor structure but with a reduced number of items (Weinman, personal communication, Moss-Morris et al. 2002). A number of items were identified as not loading clearly on to one factor and hence were excluded leaving 38 items. These versions of the IPQ-R will be referred to as IPQ-R50 and IPQ-R38 respectively. The number of items contributing to each subscale for both versions of the IPQ-R are provided in Table 6.8. The identity and causal item checklists are identical in both versions of the scale. The majority of subscales in the IPQ-R38 are shorter than those in the IPQ-R50 (timeline cyclical, consequences, personal control, treatment control, emotional representations). The timeline acute/chronic subscale is the only subscale to which an item is added in the IPQ-R38. The illness coherence subscale is identical in both versions of the scale.

Table 6.8- Number of items in subscales computed from the IPQ-R50 and IPQ-R38.

Subscale	Number of items in IPQ-R 50	Number of items in IPQ-R 38
Identity*	17	17
Timeline acute/chronic	5	6
Consequences	11	6
Personal control	9	6
Treatment control	6	5
Illness coherence*	5	5
Timeline cyclical	6	4
Emotional representations	8	6
Causal items*	19	19

*Identical items

The subscales of the IPQ-R38 were found to show good psychometric properties (Moss-Morris et al. 2002). Cronbach's alpha coefficients ranged from .79 (timeline cyclical) to .89 (timeline acute/chronic). Differing levels of stability were found across the subscales although the majority were acceptable. Test-retest reliability assessed in renal patients over 3 weeks ranged from $r = .46$, $p<.01$ (personal control) to $r = .80$, $p<.001$ (identity) and from $r = .35$ $p<0.01$ (timeline cyclical) to $r = .81$, $p<0.001$ (emotional representations) in RA patients assessed at 6 months follow-up.

6.15 AIMS AND RATIONALE

Although the IPQ and IPQ-R has shown good psychometric properties in patient populations (Weinman et al. 1996, Moss-Morris et al. 2002) the questionnaires have not previously been adapted to assess representations of healthy individuals at increased risk of illness. The applicability of the measures to women at increased risk of breast cancer is unknown. This study therefore aimed to adapt the IPQ-R to assess perceptions of breast cancer held by healthy women and assess the reliability of the measure in women at increased risk and women in the general population. This study also aimed to describe representations of breast cancer in these samples. The specific objectives for each of the aims are outlined as follows.

To adapt the IPQ-R to assess perceptions of breast cancer held by healthy women.

The IPQ-R will be described in more depth and examples of items provided. Details of how the questionnaire was adapted to this population and additional items included are provided.

To assess the reliability of using the IPQ-R to assess perceptions of breast cancer in women at increased risk and women in the general population.

The reliability of subscales derived from both versions of the IPQ-R (IPQ-R50 and IPQ-R38) will be evaluated in both the increased risk (Sample A) and general population sample (Sample B&C) in order to determine which version should be used to address the primary research question (see Chapter 4, section 4.1, pages 96-102). It was predicted that women at increased risk of breast cancer would have developed clear beliefs about the disease amenable to assessment by the IPQ-R and therefore it was expected that the subscales would exhibit adequate internal reliability in this sample. It was predicted that the IPQ-R might not perform as well in the general population sample who may not have fully formed beliefs about the disease. Test-retest reliability in the increased risk sample was expected to be higher than that reported in patient samples. Healthy individuals are not directly experiencing or responding to current symptoms or effects of illness that may cause them to reappraise and alter their perceptions of the disease in a short space of time and hence illness perceptions of healthy individuals at risk of disease were expected to be more stable over a short time frame.

To describe representations of breast cancer in women at increased risk and women in the general population.

Descriptive statistics are reported for each sample. In addition, patterns of illness perceptions are examined in each sample. Leventhal et al. (1984) conceptualised illness representations as schemata containing groups of beliefs and it is possible that patterns of illness beliefs may be more important in determining outcome than individual dimensions. Patterns of associations between the IPQ-R subscales were first explored using correlational analysis. It was predicted that intercorrelations would be comparable to that reported in patient populations (Weinman et al. 1996).

Positive associations were predicted between identity, timeline acute/chronic, consequences and emotional representations. The control subscales (treatment control and personal control) were predicted to be positively associated and to show negative correlations with timeline acute/chronic, consequences and emotional representations. Associations between the IPQ-R subscales and causal items will also be explored. This will examine associations between subscales and individual causal items as well as the overall number of causal items participants report to agree with. This was considered an important variable since previous research has shown that the number of causal attributions for illness has been associated with distress in patients and their spouses (Turnquist et al 1998, Weinman et al 2000).

Cluster analysis is a technique that has been used to assess and categorise the structure of illness representations on data derived from the IPQ (Buick et al. 1997, Moss-Morris et al. 1997, Heijmans 1999). Buick (1997) found that breast cancer patients being treated with radiotherapy could be separated into 2 clusters based on their scores on the IPQ. One cluster (named 'negative cluster') was represented by high scores on identity, timeline acute/chronic, consequences and internal blame and low scores on control/cure. Women in this cluster were more likely to report psychological distress pre and post treatment and reported greater functional disruption due to breast cancer. Heijmans (1999) reported a cluster analysis of IPQ scores from Addisons disease patients. Two clusters were found one named 'high seriousness' classified by high scores on identity, timeline acute/chronic, consequences and low scores on controllability and the other cluster named 'low seriousness' (high controllability and low identity, timeline acute/chronic and consequences). Individuals in each cluster were found to differ on levels of impairment and coping strategies.

Cluster analysis was conducted on the IPQ-R data from both samples in order to explore the patterns of representations in women at increased risk of breast cancer and women in the general population. A second objective of this analysis was to provide summaries of individual's illness perceptions in order to examine associations between patterns of illness perceptions, levels of distress and experience

of breast cancer in future analysis. It was predicted that the analysis would reveal distinct groups of individuals who differ on the IPQ-R subscales in both samples.

6.15.1 Summary of hypotheses

Table 6.9 provides an outline of the hypotheses examined in this study.

Table 6.9- Adapting the IPQ-R to assess healthy women's representations of breast cancer: Summary of hypotheses.

Aim	Hypotheses
To assess the reliability of using the IPQ-R to assess perceptions of breast cancer in women at increased risk and women in the general population.	• The IPQ-R will show adequate reliability in the increased risk sample. • The IPQ-R will show lower reliability in the general population sample. • The IPQ-R will show higher test-retest reliability in the increased risk sample than that reported in patient samples.
To describe representations of breast cancer in women at increased risk and women in the general population.	• Associations between illness perceptions will reflect that found in patient populations. • Cluster analysis will reveal distinct groups of individuals who hold different representations of breast cancer.

6.16 ADAPTING THE IPQ-R TO ASSESS HEALTHY WOMEN'S PERCEPTIONS OF BREAST CANCER.

When work began on this study only the IPQ-R50 was in the public domain. The IPQ-R50 was reworded to make it appropriate to assess healthy women's perceptions of breast cancer. Participants were asked to report their personal views about breast cancer rather than referring to their perceptions of an illness personally affecting them. For example: '*My illness has serious financial consequences'* was replaced with '*Breast cancer has serious financial consequences', 'My illness will last for a long time'* was replaced with *'Breast cancer lasts for a long time'* A brief description of each subscale with examples of items and scoring procedures are outlined in

Figure 6.1. If items are missing scores on that subscale are omitted. The full questionnaire can be seen in Appendix II (page A-9). This questionnaire is colour coded to indicate which items comprise each subscale. Details regarding reversed scoring and subscales in the shorted version of the questionnaire (IPQ-R38) are also provided.

Following previous research on perceptions of breast cancer patients (Buick 1996) and discussions with breast cancer consultants and women at increased risk, additional breast cancer specific items were generated. Three extra symptoms were added to the identity subscale ('Hard or tender growths in body', 'soreness in body', 'skin changes'). The item 'hormonal' was added to the cause subscale since oestrogen has been associated with breast cancer development (Vogel 2000). The adapted IPQ-R50 was piloted on 11 women at increased risk of breast cancer to ensure that all items were relevant and understood appropriately. These women were attending the familial breast cancer clinic in Edinburgh and were invited to provide feedback on a questionnaire designed to assess women's beliefs about breast cancer. The women were asked to read through the questionnaire and note any items they considered irrelevant or had difficulty understanding. They were also asked to record any items they felt should be included in the questionnaire. No items were consistently highlighted as problematic and there were no additional items suggested for inclusion.

Figure 6.1- IPQ-R scales assessing healthy women's beliefs about breast cancer.

Identity: Beliefs about the symptoms of breast cancer. Respondents tick the symptoms they believe to be related to breast cancer from a 17 item symptom checklist. The score is the total number of symptoms ticked.

The items for the following 7 subscales are presented in a random order. Responses are rated on a five point scale from 'strongly disagree' to 'strongly agree'. Subscale scores are the mean of items (after reverse scoring as necessary).

Timeline acute/chronic: Beliefs about the duration of breast cancer. 5 items. Higher scores indicate a perception of breast cancer as more chronic. (Eg. '*Breast cancer lasts for a lifetime'.)*

Consequences: Beliefs about the impact of breast cancer on everyday life. 11 items. Higher score represents perceptions of breast cancer as holding more serious consequences (Eg. '*Breast cancer has major consequences on patients' lives*').

Personal control: 9 items. Higher score indicates a greater perception of personal control over breast cancer. (Eg '*Patients have the power to influence breast cancer*').

Treatment control: Beliefs about the efficacy of treatment to cure and control breast cancer. 6 items. Higher score indicates greater perceived efficacy of treatment. (Eg '*Treatment is effective in curing breast cancer'*).

Illness coherence: The extent to which the participant's perceptions of breast cancer are coherent. 5 items. Higher score indicates a less coherent perception of breast cancer. (Eg '*Breast cancer doesn't make any sense to me'*).

Timeline cyclical: Beliefs about the prognosis of breast cancer. 6 items. Higher score indicates a perception of breast cancer as more variable and unpredictable. (Eg '*Breast cancer goes through cycles in which it gets better and worse'*).

Emotional representations: Emotional responses to breast cancer. 8 items. Higher score represents greater emotional response to breast cancer. (Eg '*When I think about breast cancer I get upset'*).

Cause: Respondents endorse 19 statements about the causes of breast cancer on a 5 point scale ranging from 'strongly disagree' to 'strongly agree'. These items are retained individually. Respondents are also asked to state what they believe to be the 3 most important causes of breast cancer.

6.17 DESIGN AND SAMPLE

The study was a cross sectional questionnaire study. The design and procedure were described in Chapter 4. Data reported in this Chapter were derived from the following samples: Increased risk sample (Sample A) (n= 117) and general population sample (Sample B&C)(n= 256). Test-retest data are also examined in the follow-up sample of at risk women (n= 74). These samples were described in Chapter 5 (see sections 5.2 and 5.3, pages116- 119).

6.18 STATISTICAL ANALYSIS

6.18.1 Reliability analysis

Reliability analysis was conducted on both versions of the IPQ-R (IPQ-R50 and IPQ-R38) in order to determine which version of the IPQ-R to use in subsequent analysis. Internal consistency of the IPQ-R subscales were assessed using Cronbach's alpha. Test-retest reliability was obtained in the follow-up sample and examined using Pearson's correlation coefficient.

6.18.2 Descriptive statistics

Histograms, boxplots and normal probability plots were used to examine the distribution of subscales including the symmetry of distribution, spread of scores and outliers. Descriptive statistics including mean, skew and kurtosis were also obtained and examined for problems in the data.

6.18.3 Patterns of illness perceptions

6.18.3a Intercorrelations

In both samples correlations were examined between the IPQ-R subscales. Exploratory analysis was also conducted to examine correlations between IPQ-R subscales and causal items and between the causal items. Due to the number of correlational analysis conducted significance levels of $p<0.01$ are reported here.

6.18.3b Cluster analysis

6.18.3.b(i)Background to cluster analysis

Cluster analysis was used to assess the patterns of illness representations in both samples. This is a descriptive, exploratory technique that searches for characteristic patterns in scores across a number of variables. It combines individuals into groups or 'clusters' so that individuals in the same cluster are more alike (with respect to the variables in question) than they are to individuals in another cluster. Cluster analysis uses heuristics and plausible algorithms to create clusters and there are no statistical assumptions for this technique. There are many different methods available based upon a variety of algorithms that aim to maximise the differences between clusters relative to the variance within the clusters. There are two broad categories of methods: Hierarchical and non hierarchical.

Hierarchical methods classify individuals into clusters in a series of stages in which progressively larger groups are formed by joining earlier clusters. Agglomerative hierarchical methods start this process with each individual in their own cluster and progress until all individuals are in the same cluster. There are a number of different techniques for these methods based on different ways of defining distance between individuals and clusters in order to determine cluster membership. The hierarchical stages of these methods are represented graphically by dendrograms that illustrate the fusions/divisions at each stage (please see Figure 6.2). The dendrogram is examined in order to determine the optimum clusters that represent the data. There are no standard selection procedures and judgements are made on a subjective interpretation of changes in the dendrogram (Everitt 1993). It has been suggested that researchers examine a number of cluster solutions in light of theoretical expectations (Hair et al. 1995).

Non-hierarchical (K means) cluster analysis is conducted by specifying the number of clusters expected in the sample. The researcher selects cluster centres (cluster seeds) and individuals are subsequently assigned to one of the clusters. The methods utilised in this approach differ on techniques used to obtain cluster seeds and rules for assigning individuals into clusters (Sharma 1996).

Both hierarchical and non-hierarchical methods have advantages and disadvantages and can produce different cluster solutions of the same data set. Hierarchical methods are affected by outliers and are difficult to compute on large datasets. Non-hierarchical methods are less susceptible to outliers but depend on the researcher to select appropriate cluster seeds according to theoretical or practical considerations. Different seeds can produce very different cluster solutions. Hair et al. (1995) reports that non-hierarchical methods are superior to hierarchical methods when cluster seeds can be specified. Sharma (1996) and Hair et al. (1995) have suggested that combining the approaches maximises the advantages of both techniques and allows the researcher to be most confident in the validity of the solution. Hair et al. (1995) suggests combining the approaches allows non-hierarchical techniques to 'fine tune' the results from hierarchical analysis by allowing individuals to change cluster membership. This method will be utilised on the IPQ-R data set from both samples and is outlined below (6.18.3b(ii)).

Once the cluster solution is obtained clusters must be interpreted and assigned a name that reflects the nature of the cluster. It is important to determine the variables that differentiate the clusters and the patterns of response within each cluster. Since cluster solutions will differ depending on what method is utilised it is important to validate the solution in order to determine that the clusters are real and not merely imposed on the data by the method. There are a number of ways to validate a cluster solution. For example, performing the analysis on a separate sample and comparing the cluster solutions and cluster membership and testing the clusters for differences expected on other variables.

Although cluster analysis does not incorporate any statistical assumptions it is important to consider the variables entered into the analysis. The selection of variables for analysis should be theoretically driven since cluster analysis is unable to distinguish relevant and irrelevant variables. The distance measures are also sensitive to differing scales in that scales with larger dispersion have more impact on the solution. Standardized score (z scores) are used in order to eliminate this bias.

6.18.3b (ii) Cluster analysis of IPQ-R data

Step 1.
Standardized scores (z scores) of the IPQ-R subscales were calculated for each participant for use in analysis (Identity; timeline acute/chronic; timeline cyclical; consequences; treatment control; personal control; illness coherence; emotional representations). Subscales that showed adequate psychometric properties were included in the analysis. The causal items were not included in the analysis since there were may single items and no clear predictions as to how these items would cluster.

Step 2.
A number of hierarchical agglomerative cluster analysis were performed on the data from each sample (Wards method, centroid method, single linkage and complete linkage) using squared Euclidean distance as the similarity measure. These are the most widely used methods and distance measures in empirical studies (Everitt 1993, Everitt 1996). Dendrograms for these analyses were examined in order to determine the number of clusters in each sample. Means for each variable from the clusters from the Wards method were obtained for use in the non-hierarchical analysis. The Wards method is considered the best hierarchical method available (Hair et al. 1995, Everitt 1996, Hack and Degner 1999).

Step 3.
A non-hierarchical Kmeans cluster analysis was performed specifying the number of clusters identified by the hierarchical analysis and using the means for each variable in each cluster as seed points. Clusters were interpreted and named by comparing means scores on each of the unstandardized IPQ-R subscales.

Step 4.
To validate the cluster solution another Kmeans cluster analysis using random cluster seed points was obtained. The results were compared in terms of cluster sizes and cluster membership. The cluster solutions obtained for both samples were also compared. Further validation of the cluster solutions will be explored throughout the thesis by examination of predicted associations between illness perception clusters, psychological well-being and experience of breast cancer in the family.

6.19 RESULTS

6.19.1 Reliability analysis

6.19.1a Internal consistency

The alpha coefficients for both versions of the IPQ-R in each sample are provided in Table 6.10. In the increased risk sample (A) both versions of the emotional representations, treatment control, personal control and illness coherence subscales performed well (Cronbach's alpha >.7). The identity, timeline acute/chronic and consequences subscales were marginal in this sample (>0.55). The longer versions of these subscales (derived from the IPQ-R50) showed higher levels of internal consistency than those from the shorter version (IPQ-R38). Both versions of the timeline cyclical subscale showed extremely low internal consistency.

In the general population sample (B&C) the identity, emotional representations and illness coherence subscales showed acceptable internal consistency (>.7). The longer version of the consequences subscale performed well (.76) but the shorter version showed reduced internal consistency (.66). Both versions of the timeline acute/chronic subscale, personal control and treatment control subscales were marginal. The longer versions of the timeline acute/chronic subscale and personal control subscales performed slightly better than the short versions. Both versions of the timeline cyclical subscales showed very poor internal consistency.

Comparing the internal consistency of the subscales derived from the IPQ-R between the increased risk (A) and general population sample (B&C) suggested that the emotional representations and illness coherence subscales performed well across all samples. The identity and consequences subscales showed higher internal consistency in the general population sample than the increased risk sample. Both the personal and treatment control subscales showed higher levels of internal consistency in the increased risk sample than the general population sample. The timeline acute/chronic subscales was marginal in both samples and the timeline cyclical subscale performed poorly in all samples.

Cronbach's alpha was systematically re-calculated for each subscale with each item omitted in order to identify problematic items. Item deletion was found to only marginally improve the consistency of a few subscales. The subscales that could be improved by omitting items are highlighted with a star (*) in Table 6.10. This analysis is reported in Appendix III (page A-21). The largest improvement was in the general population sample in which the 38-item version of the timeline cyclical subscale was improved from .39 to .49 with item 4 deleted. However this is still an unacceptable level of internal consistency.

Table 6.10-- Internal consistency (Cronbach's Alpha) for IPQ-R50 and IPQ-R38 subscales in the increased risk and general population samples

Sample	Increased risk sample (A)		Full general population sample (B&C)	
IPQ-R subscale	IPQ-R50	IPQ-R38	IPQ-R50	IPQ-R38
Identity	.66*	-	.77*	-
Timeline acute/chronic	.61	.59*	.63*	.61*
Consequences	.69*	.56*	.76	.66
Personal control	.82*	.78*	.68	.65
Treatment control	.72*	.73*	.60*	.63*
Illness coherence	.81	-	.84	-
Timeline cyclical	.35*	.43	.31*	.39*
Emotional representations	.89	.87	.87*	.85

6.19.1b Test re-test reliability

Test re-test reliability was calculated for the follow up subsample of the increased risk sample (see 5.2.1a (page 116) and Figure 5.1 (page 118) for details of sample). The correlation coefficients for both versions of the subscales are provided in Table 6.11. The test-retest values are high for both versions of the subscales. All correlation coefficients for the full sample are significant ($p<0.001$) except the longer version of the timeline cyclical ($p=0.006$). Test retest reliability for individual items comprising the timeline cyclical subscale were assessed in order to explore why stability was lower in this scale. Five of the six items showed significant test retest reliability

ranging from r= .34, p= 0.003, n= 73 (item 4) to r= .58, p<0.001, n= 72 (item 34). One item (item number 7 *'Patients experience breast cancer symptoms pretty much all of the time'*) did not show a significant correlation coefficient (r= . 04, p= .74, n= 73). This suggests that this item in particular is not reliable in this population.

Test-retest correlation coefficients were also computed for participants who did (n= 37, 43%) and did not (n= 42, 57%) report experiences that may have influenced their thoughts and feelings about breast cancer between the questionnaires. These results are fully reported in Appendix III (page A-22). The correlation coefficients are generally higher for participants who did not report breast cancer related experiences between questionnaires than for participants who did. The test re-test value for the timeline cyclical subscale was non significant for participants who reported a breast cancer related experience between the questionnaires (r=-.2, p>.05).

Table 6.11- Test-retest correlation coefficients for all respondents in the follow-up sample.

Subscale	IPQ-R50		IPQ-R38	
	n	r	n	r
Identity	74	.54***	-	-
[3]Timeline acute/chronic	67	.68***	66	.66***
Consequences	68	.77***	70	.72***
Personal control	68	.74***	68	.74***
Treatment control	68	.77***	69	.74***
Illness coherence	70	.68***	-	-
Timeline cyclical	70	.33**	70	.43***
Emotional representations	64	.88***	64	.85***

***p<0.001

**p<0.01

Since the majority of the subscales derived from the IPQ-R50 showed greater internal consistency than those derived from the IPQ-R38 in both samples and higher test-retest reliability in the increased risk sample it was decided to utilise this version

[3] The sample size is higher in the IPQ-R50 version the timeline acute/chronic scale because this version is shorter than that derived from the IPQ-R38 (see section 6.14, page 159).

of the questionnaire in subsequent analysis. Following reference to the IPQ-R refers to this version of the measure.

6.19.2 Descriptive statistics

6.19.2a Increased risk sample (A)

One hundred and seventeen women in the increased risk sample completed the IPQ-R. Two participants failed to complete page 9 of the questionnaire and 2 other participants failed to complete page 10 of the questionnaire. Both of these pages contained items from the consequences, timeline acute/chronic, personal control, treatment control, illness coherence and emotional subscales. Since scores are not derived for subscales with missing items the maximum sample size for these subscales was reduced to 113. Page 9 contained items from the timeline cyclical subscales and hence the maximum sample size for this subscale was reduced to 115. Other missing data appeared random with no item omitted more than twice. The sample size for the subscales ranges from 105 (emotional representation) to 117 (identity).

Table 6.12 provides the descriptive statistics of the IPQ-R subscales. Examination of graphical distribution and descriptive statistics did not reveal any problems in the data. The identity subscale showed a slight left (positive) skew and the consequences, treatment control and emotional representations subscales showed a slight right (negative) skew. No subscale had more than 4 outlying cases. The questionnaire ID number was used to identify questionnaires that included outliers and checks were made to ensure there were no reasons for these values (eg mistakes in data input or written explanations on questionnaires). No evidence was found to suggest that those participants who had outlying scores were unrepresentative and there was not sufficient justification for removing these scores (Altman 1991).

Table 6.12 - Descriptive statistics of the IPQ-R subscales in the increased risk sample (A).[4]

IPQ-R subscale	n	Mean	sd
Identity	117	4.4	2.25
Timeline acute/chronic	111	3.3	0.53
Consequences	109	3.9	0.38
Personal control	111	3.3	0.60
Treatment control	112	3.6	0.52
Illness coherence	111	2.5	0.69
Timeline cyclical	113	3.1	0.42
Emotional representations	105	3.3	0.69

Causal items

On average women in the increased risk sample agreed with 6.2 (sd 3.20) of the cause items (rated agree/strongly agree). The rating scores for each item can be seen in Table 6.14. The highest rated items (with mean values over 3) were: heredity, hormonal, smoking, diet, ageing, stress, and chance. When asked to state the three most important factors believed to cause breast cancer 113 (97%) endorsed heredity; 49 (42%) hormonal factors; 37 (32%) diet; 34 (29%) smoking; 26 (22%) ageing; 23 (20%) stress or worry; 17 (15%) a germ or virus. Only 4 other causes were noted which were not already provided. Two of these were similar to causes already on the list ('natural mutation' and 'random') two others were new causes not on the IPQ-R list ('under wired bras' and 'persons body').

6.19.2b General population sample (B&C)

Missing data in this sample also appeared random. Sample size for the subscales ranged from 237 (personal control) to 256 (identity). Table 6.13 provides the descriptive statistics for IPQ-R subscales. The majority of subscales in this sample appeared normally distributed. The identity subscale showed a slight left (positive) skew and the treatment control subscale showed a slight right (negative) skew. The timeline cyclical subscale showed a high numbers of outliers (n= 21). Checks were made to ensure that participants with outlying scores did not differ from the

[4] Scores are computed by summing items and obtaining mean (see section 6.14, page 159-160)

remaining sample in terms of background demographics. No differences were found and there were no suggestions that these participants were unrepresentative.

Table 6.13- Descriptive statistics of the IPQ-R subscales in the general population sample (B&C)

IPQ-R subscale	N	Mean	Sd
Identity	256	5.1	2.86
Timeline acute/chronic	243	3.2	0.53
Consequences	240	3.9	0.44
Personal control	237	3.2	0.47
Treatment control	244	3.6	0.42
Illness coherence	245	3.1	0.38
Timeline cyclical	238	2.9	0.70
Emotional representations	240	3.1	0.68

Causal items

On average women in the general population sample agreed with 6 (sd 3.41) of the cause items (rated agree/strongly agree). The rating scores for each item can be seen in Table 6.14. The highest rated items with mean values over 3 were heredity, hormonal factors, smoking, ageing, stress, chance and diet. When asked to rank the three most important factors believed to be causes of breast cancer 226 (88%) endorsed heredity; 105 (41%) hormonal factors; 93 (36%) smoking; 53 (21%) stress; 53 (21%) chance or bad luck; 42 (16%) ageing; 27 (11%) diet. Altogether 36 different causes not provided by the IPQ-R50 were recorded. Twenty of these causes overlapped with those already on the list. The 16 different causes reported were: 'over exposure to the sun', 'medication', 'problem with breasts', 'lump in breast', 'contraceptive pill', 'not having children', 'fatty tissue in breast', 'exposure to carcinogenic factors', 'menopause', 'age at first pregnancy',' too many X rays', 'never having breast fed', 'hard growths in body', 'soreness', 'abortion' and 'modern living (food, radiation, radiowaves)'.

Table 6.14- Descriptive statistics of causal items in the increased risk (A) and general population sample (B&C)

	Increased risk sample (A)			General population sample (B&C)		
Causal items:	n	Mean	sd	n	Mean	sd
Stress or worry	116	3.2	0.99	251	3.2	0.93
Heredity - it runs in the family	117	4.6	0.50	255	4.4	0.64
A germ or virus	116	2.1	0.93	246	2.3	0.84
Diet or eating habits	117	3.3	0.99	249	3.1	0.96
Chance or bad luck	117	3.0	1.12	249	3.2	1.11
Poor medical care in the past	117	2.3	0.92	248	2.5	0.84
Pollution in the environment	116	3.0	0.91	252	3.0	0.90
Patients own behaviour	116	2.5	0.95	249	2.6	0.86
Patients mental attitude e.g. thinking about life negatively	117	2.5	0.93	249	2.6	0.87
Family problems or worries causes breast cancer	115	2.5	0.87	250	2.6	0.88
Overwork	117	2.4	0.75	249	2.4	0.74
Emotional state e.g. feeling down, lonely, anxious, empty	116	2.5	0.85	250	2.6	0.88
Ageing	116	3.2	0.93	252	3.2	0.93
Alcohol	116	2.7	0.91	246	2.7	0.83
Smoking	115	3.4	0.99	252	3.5	0.88
Accident or injury	115	2.6	0.95	248	2.7	0.93
Patient's personality	117	2.2	0.78	248	2.2	0.73
Altered immunity	117	2.9	1.05	248	3.0	0.88
Hormonal	117	3.7	0.75	252	3.6	0.78

6.19.3 Patterns of beliefs[5]

6.19.3a Intercorrelations

6.19.3a(i) Increased risk sample (A)

Intercorrelations between IPQ-R subscales ($p<0.1$) are provided in Table 6.15. As predicted, the identity, timeline acute/chronic, consequences and emotional

[5] Timeline cyclical subscale is omitted from this analysis following poor internal consistency and reliability (see section 7.13.1).

representations subscales showed positive correlations, and the control subscales (personal and treatment control) were significantly positively correlated. These results suggest that women perceiving a strong identity for breast cancer were more likely to perceive the illness as long-lasting with severe consequences ($p<0.05$). Individuals who perceived more personal control over breast cancer were more likely to perceive treatment as effective ($p<0.01$). Individuals with stronger emotional representations of breast cancer perceived breast cancer as long-lasting, with severe consequences. Individuals with less belief in the treatment control of breast cancer were more likely to perceive the illness as more long-lasting and chronic and with more severe consequences ($p<0.05$). The predicted association between emotional representations of breast cancer and control beliefs were not observed in this sample.

The new illness coherence subscale showed intercorrelations with other subscales. Individuals scoring higher on the illness coherence subscale (who did not have a coherent understanding of the disease) were more likely to score higher on the identity subscale and to have shown diminished perceptions of personal control of breast cancer ($p<0.05$).

Exploratory analysis of associations with causal item revealed that the number of causes participants agreed with was significantly correlated with consequences ($r= .32$, $p= <0.001$, $n= 109$), personal control ($r= .23$, $p= 0.015$, $n= 111$) and illness coherence ($r= -.19$, $p= 0.044$, $n= 111$). Therefore participants who believed in more causes of breast cancer believed the disease to be hold greater consequences were more likely to believe in the personal control of the disease and held a more coherent understanding of breast cancer. A number of correlations were found between the IPQ-R subscales and causal items. Due to the number of tests conducted the p value was reduced to $p<0.01$. The significant results are reported in Table 6.17. The consequences and the personal control subscales showed the greatest number of correlations with causal items. There were also a large number of intercorrelations within the causal items in each sample. In the increased risk sample there were 66 significant correlations ($p<0.01$) between causal items.

Table 6.15-Pearson's correlation coefficients between IPQ-R subscales in the increased risk sample (A) (p<0.1 reported).

IPQ-R subscale	Identity	Timeline acute/ chronic	Consequences	Personal control	Treatment control	Illness coherence
Timeline acute/chronic	r= .20 p= 0.04 n= 111					
Consequences	r= .22 p= 0.02 n= 109	r= .36 p< 0.001 n= 107				
Treatment control		r = .-41 p< 0.001 n= 110	r=-.20 p= 0.035 n= 108	r= .44 p< 0.001 n =110		
Illness coherence	r= .21 p= 0.028 n= 111	r = .17 p= 0.071 n= 109		r= .-27 p= 0.004 n=109		
Emotional representation	r= .173 p= 0.078 n= 105	r= .23 p= 0.019 n=103	r= .30 p= 0.002 n=103			r= .18 p= 0.071 n= 104

6.19.3a(ii) General population sample (B&C)

The general population sample showed a similar pattern of intercorrelations to the increased risk sample. The correlation matrix is provided in Table 6.16. The consequences subscale was significantly positively correlated with identity and timeline acute/chronic as predicted, although timeline acute/chronic was not associated with identity ($p<0.05$). Emotional representations subscale was significantly associated with all other subscales in the expected direction (positively associated with identity, timeline acute/chronic, consequences and negatively associated with personal and treatment control). Women who had a stronger emotional response to breast cancer were less likely to believe in the efficacy of treatment in controlling breast cancer or personal control over the disease ($p<0.05$). Both treatment control and personal control were negatively associated with timeline acute/chronic, as predicted ($p<0.05$). Women who believed in the control of breast cancer were less likely to believe the disease was long-lasting ($p<0.001$). However

neither control subscale were associated with identity or consequences as was predicted.

In this sample the illness coherence subscale was negatively correlated with treatment control ($p<0.05$). Women with a stronger belief in the efficacy of treatment to control breast cancer had a more coherent understanding of the disease.

Exploratory correlational analysis of the causal items revealed that the number of causes participants agreed with was correlated with a number of subscales (consequences $r= .28$, $p< 0.001$, $n= 240$ and personal control $r= .16$, $p= 0.017$, $n= 237$). In addition, participants in the general population who reported to believe in more causes of breast cancer were more likely associate a greater number of symptoms with the disease (identity $r= .25$, $p< 0.001$, $n= 256$) and to have a stronger emotional response to breast cancer ($r= .20$, $p= 0.002$, $n= 240$). A number of significant correlations were found between causal items and the other belief subscales ($p<0.01$). These are reported in Table 6.17. Generally there were more correlations between the causal items and belief subscales in the general population sample compared to the increased risk sample, however the correlation coefficients were not particularly high. In the general population sample there were 108 significant correlations between causal items ($p<0.01$).

Table 6.16-Pearson's correlation coefficients between IPQ-R subscales in the general population sample (B&C) (p<0.1 reported).

IPQ-R subscale	Identity	Timeline acute/ chronic	Consequences	Personal control	Treatment control	Illness coherence
Consequences	r= .13 p= 0.05 n= 240	r= .33 p< 0.001 n= 235				
Personal control		r= -.27 p< 0.001 n= 234				
Treatment Control		r= -.31 p<0.00 n= 239		r= .40 p< 0.001 n= 233		
Illness coherence	r= -.11 p= 0.09 n= 245			r= -.12 p= 0.06 n= 233	r= -.14 p= 0.03 n= 240	
Emotional representation	r= .19 p= 0.004 n= 240	r= .30 p< 0.001 n= 235	r= .28 p< 0.001 n= 233	r= -.15 p= 0.02 n= 228	r= -.15 p= 0.02 n= 236	r= .13 p= 0.05 n= 237

Table 6.17- Pearson's correlation coefficients between IPQ-R subscales and causal items in both samples ($p<0.01$ reported).

Subscale	Increased risk sample (A)	General population sample (B&C)
Identity	Accident or injury (r=.31, n= 115, p= 0.001) Smoking (r=.28, n= 115, p= 0.003)	Heredity (r=.23, n= 255, p=0.000) Emotional state (r=.22, n= 250, p=0.001) Family problems (r=.19, n= 250, p=0.003) Stress (r=.18, n= 251, p=0.004)
Timeline acute/chronic		Accident or injury (r=.19, n= 236, p=0.004)
Consequences	Altered immunity (r=.33, n= 109, p=0.000) Patients personality (r=.32, n= 109, p=0.001) Patient mental attitude (r=.31, n= 109, p=0.001) Emotional state (r=.29, n= 108, p= 0.002) Chance (r=.29, n= 109, p=0.003) Stress (r=.27, n= 108, p=0.005)	Heredity (r=.31, n= 240, p=0.000) Hormonal (r=.30, n= 237, p=0.000) Overwork (r=.27, n= 234, p=0.000) Emotional state (r=.26, n=235, p=0.000) Family problems (r=.25, n=235, p=0.000) Smoking (r=.20, n=237, p=0.002) Patient mental attitude (r=.20, n=234, p=0.001) Altered immunity (r=.18, n=232, p=0.006) Ageing (r=.18, n=237, p=0.006) Alcohol (r=.17, n=231, p=0.001)
Personal control	Patients mental attitude (r=.36, n= 111, p=0.000) Diet (r=.34, n= 111, p=0.000) Patients behaviour (r=.32, n= 111, 0.000)	Patients behaviour (r=.28, n=231, p=0.000) Patients personality (r=.24, n=230, p=0.000) Pollution (r=.20, n=234, p=0.002) Patients mental attitude (r=.20, n=231, p=0.003) Accident or injury (r=.-18, n=230, p=0.007) Stress or worry (r=.17, n=233, p=0.009) Diet or eating habits (r=.17, n=232, p=0.009)
Illness coherence		Germ or virus (r=.21, n=237, p=0.001)
Emotional representations		Overwork (r=.22, n=234, p=0.001) Accident or injury (r=.22, n= 233, p=0.001) Hormonal (r=.20, n= 237, p=0.002)

		Emotional state (r=.20, n= 235, p=0.002) Family problems (r=.19, n=235, p=0.004) Stress (r=.17, n= 235, p=0.001)

6.19.3b Cluster analysis[6]

6.19.3b(i) Increased risk sample (A)

The dendrograms from Wards hierarchical analysis clearly suggested a 2 cluster solution (see Figure 6.2). The cluster means for these clusters on each of the variables were obtained and entered as the cluster seed points for the non-hierarchical analysis.

The cluster solution obtained from the non-hierarchical analysis showed a similar number of participants in each cluster. Cluster 1 contained 51 (52%) participants and cluster 2 contained 47 (48%) participants. As Table 6.18 indicates the clusters differed significantly on all the IPQ-R subscales ($p<0.05$). Participants in cluster 1 believed breast cancer: to have more symptoms; to be longer lasting and hold greater consequences. They also held stronger emotional representations of breast cancer than participants in cluster 2. Those in cluster 1 were less likely to believe in treatment or personal control of breast cancer and held a less coherent understanding of the disease those participants in cluster 2. The clusters seem to represent a dichotomy of beliefs and were thus labelled 'negative representation' (cluster 1) and 'positive representation' (cluster 2) respectively. The mean z scores on the IPQ-R subscales for each cluster are demonstrated graphically in Figure 6.3.

In order to validate this analysis another K-means cluster analysis was conducted using random cluster seed points and the results compared. The cluster solution was similar. Each cluster differed significantly on all of the IPQ-R subscales and the means on each subscale were comparable to those found in the previous solution. The sizes of the clusters were also comparable ('negative representation' cluster n=

[6] Timeline cyclical subscale is omitted from this analysis following poor internal consistency and reliability (see section 6.19.1, page 171-174).

55; 'positive representation' cluster n= 43). There were only 5 differences in classification of participants compared with the previous solution. One woman who was classified in the negative cluster previously was in the positive cluster in the analysis using random cluster seed points. Four women classified in the positive cluster in the previous analysis were found in the negative cluster in the random seed analysis. The comparability of cluster solutions indicates that the clusters reliably differentiate participants on the basis of scores on the IPQ-R.

Table 6.18- Cluster analysis of IPQ-R subscales in the increased risk sample (A).

IPQ-R subscale Mean, (sd)	Cluster 1 'negative representation' (n=51)	Cluster 2 'positive representation' (n=47)	t	df	p
Identity	5.4 (2.42)	3.5 (1.41)	4.61	96	.000
Timeline acute/chronic	3.5 (0.51)	3.0 (0.46)	4.46	96	.000
Consequences	4.0 (0.31)	3.8 (0.42)	2.81	96	.006
Personal control	2.9 (0.56)	3.6 (0.48)	-6.11	96	.000
Treatment control	3.4 (0.53)	3.9 (0.36)	-5.69	96	.000
Illness coherence	2.8 (0.62)	2.1 (0.53)	6.07	96	.000
Emotional representation	3.6 (0.55)	3.0 (0.74)	4.29	96	.000

Figure 6.2- Dendrogram from hierarchical cluster analysis of IPQ-R subscales(Wards method) in the increased risk sample (A).

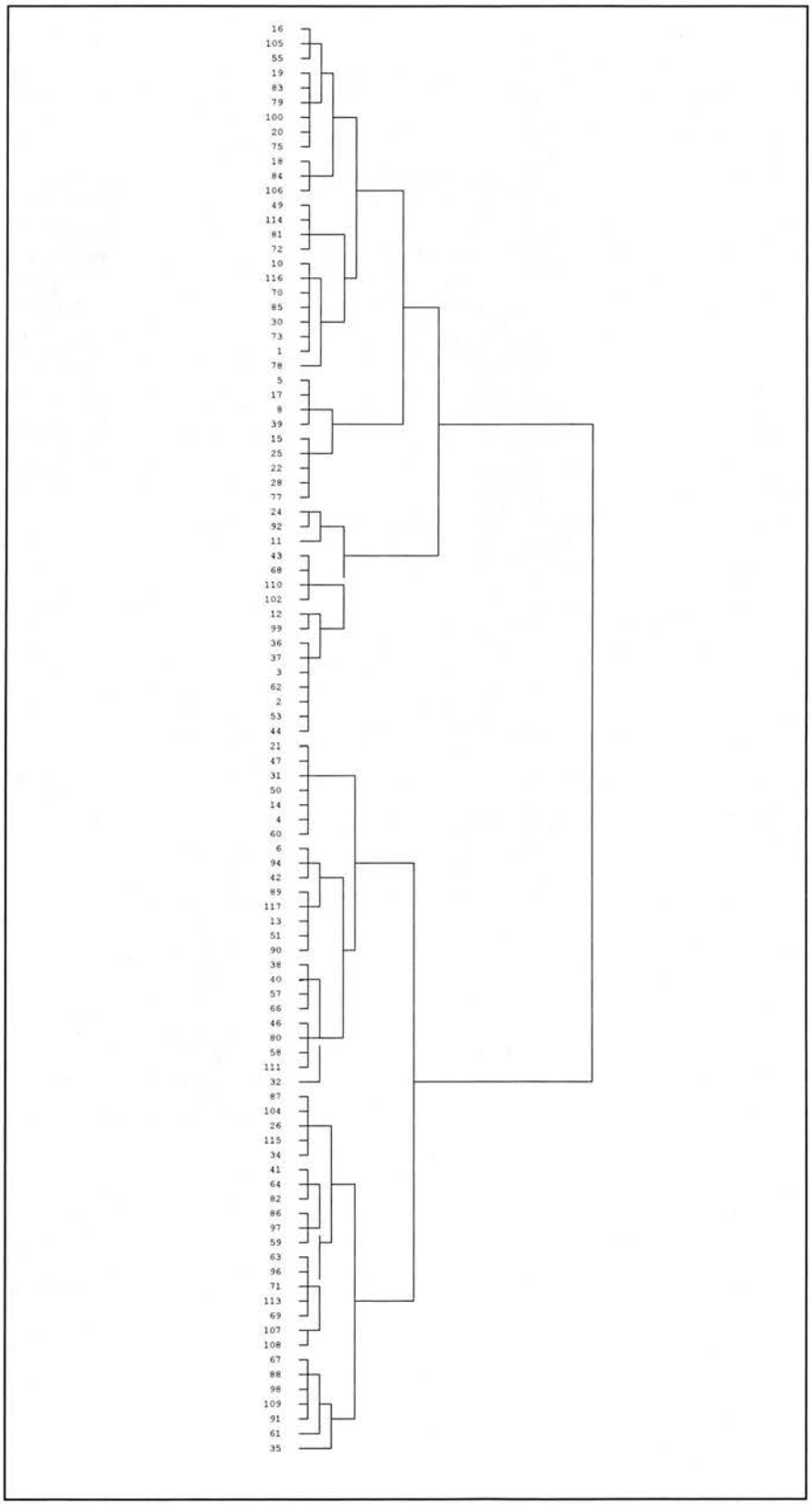
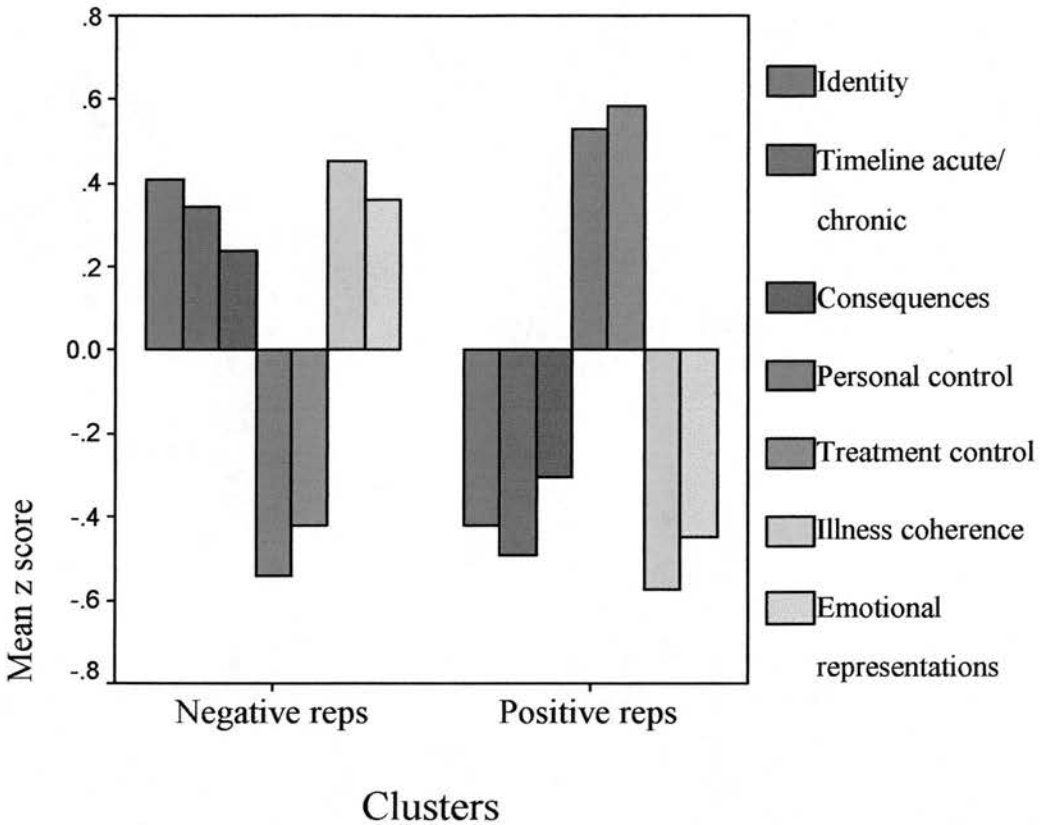


Figure 6.3- Mean z scores on the IPQ-R subscales for each cluster in the increased risk sample (A).



Exploratory analysis was conducted to assess differences between the clusters on causal items. There was no significant difference in the number of causes believed to be associated with breast cancer between participants in the positive and negative clusters (t=-1.53, df= 96, p= 0.129). However, participants in the positive cluster were more likely to agree that 'family problems', 'overwork' and 'emotional state' are causes of breast cancer than those in the negative cluster ($p<0.05$). There was no significant difference between the clusters on age or risk perception ($p<0.05$). There was a trend for individuals in the negative cluster to report slightly higher perceptions of risk (mean = 3.1, sd= 0.72) than individuals in the positive cluster (mean= 2.8, sd= 0.77) (t=1.67, df= 95, p= 0.098).

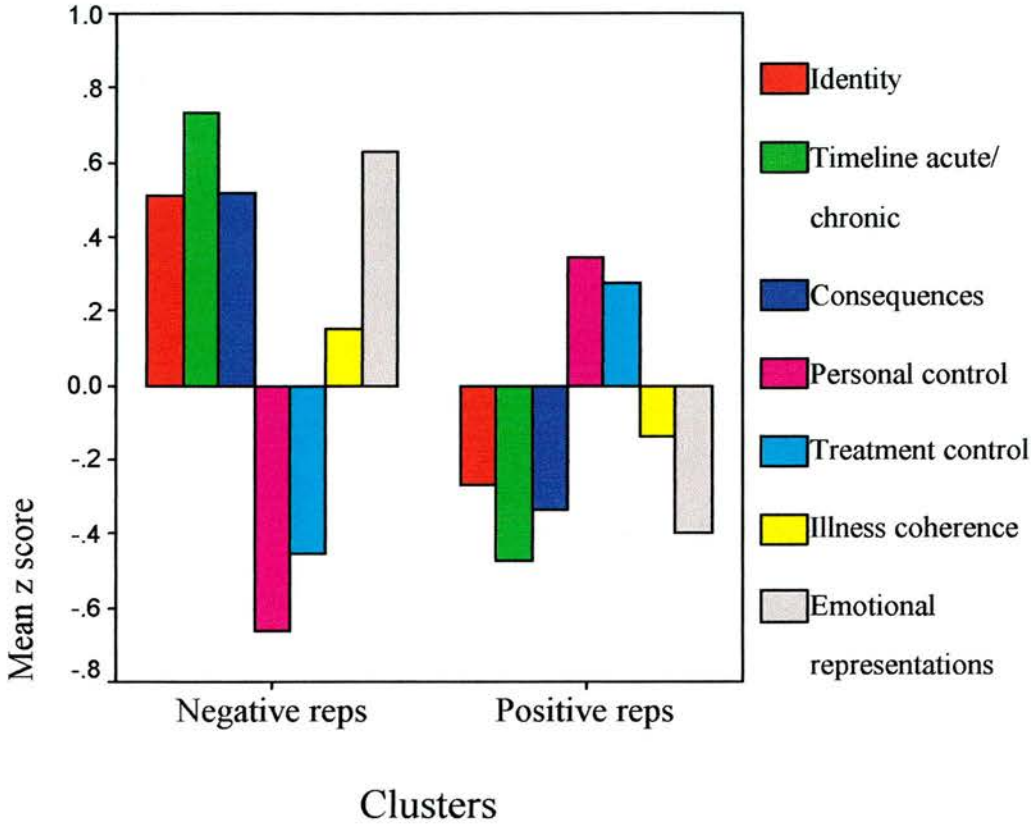
6.19.3b(ii) General population sample (B&C)

The dendrogram from the Wards hierarchical cluster analysis also suggested a 2 cluster solution in the general population (see Appendix III page A-23). When the means were entered into a K-means cluster analysis the cluster solution observed reflected that found in the increased risk sample. The clusters differed significantly on all of the IPQ-R subscales ($p<0.05$) (Please see Table 6.19). Cluster 1 ('negative representation') were defined by high scores on the identity, timeline acute/chronic, consequences and emotional representations subscales and low scores on the personal and treatment control subscales. The mean z scores on the IPQ-R subscales for each cluster are demonstrated graphically in Figure 6.4. The distribution of participants between the clusters was different in the general population sample than the increased risk sample. The 'negative representation' cluster encompassed 36% of participants and the 'positive representation' cluster held 64% of participants. When the random K-means cluster analysis was computed exactly the same cluster solution was obtained with the same participants in each cluster suggesting that the solution obtained was reliable.

Table 6.19- Cluster analysis of IPQ-R subscales in the general population sample (B&C).

IPQ-R subscale Mean, (sd)	Cluster 1 'negative representation' (n=78)	Cluster 2 'positive representation' (n=138)	t	df	p
Identity	6.5 (3.46)	4.3 (2.20)	5.79	241	.000
Timeline acute/chronic	3.6 (0.47)	3.0 (0.38)	10.72	214	.000
Consequences	4.1 (0.37)	3.8 (0.41)	6.68	214	.000
Personal control	2.9 (0.46)	3.4 (0.39)	8.01	214	.000
Treatment control	3.5 (0.45)	3.8 (0.34)	5.62	214	.000
Illness coherence	3.0 (0.69)	2.8 (0.69)	2.07	214	.040
Emotional representation	3.6 (0.56)	2.8 (0.61)	8.38	214	.000

Figure 6.4- Mean z scores on the IPQ-R subscales for each cluster in the general population sample (B&C).



Exploratory analysis revealed that the clusters differed on a number of the causal items. Participants in the negative cluster reported to believe in on average 6.9 causes of breast cancer compared to 5.7 of those in the positive cluster (t= 2.39, df= 214, p= 0.018). Participants in the negative cluster were significantly more likely to agree that 'heredity'; 'chance or bad luck'; 'overwork'; 'emotional state'; 'accident or injury' and 'hormonal factors' were causes of breast cancer ($p<0.05$). There were no significant differences in age or risk perception between the cluster groups. As in the increased risk sample there was a trend for individuals in the negative cluster to report slightly higher perception of risk (mean =2.5, sd= 0.72) than those in the positive cluster (mean =2.3, sd= 0.57) (t= 1.93, df= 212, p= 0.056).

6.20 SUMMARY AND DISCUSSION

6.20.1 Adaptation of the IPQ-R to assess perceptions of breast cancer held by healthy women.

The IPQ-R was easily adapted to assess perceptions of breast cancer in healthy women. A few breast cancer specific items were added to the identity and cause checklists and pilot interviews suggested that the item were relevant and comprehensible to women with a family history of breast cancer. Results from the cross-sectional study revealed that the distribution of missing items was random and did not suggest that women were having trouble responding to any particular item or subscale. Missing data for any subscale in either sample did not exceed 10%. This proportion of missing data is common in questionnaires following illegible, invalid or omitted responses (Streiner and Norman 1991).

6.20.2 Reliability of the IPQ-R to assess perceptions of breast cancer held by healthy women.

The reliability of both versions of the IPQ-R (IPQ-R50 and IPQ-R38) were examined in both the increased risk and general population samples. The internal consistency and test re-test reliability of the subscales derived from the IPQ-R50 were generally higher than the subscales calculated from the IPQ-R38. This suggests that the longer version of the questionnaire is more appropriate in these samples. It is possible however that the Cronbach alpha value for the longer version (IPQ-R50) may have been inflated due to the increased number of items. Cronbach alpha is a function of *both* the average inter-item correlation and the number of items. The use of general guidelines for interpreting this statistic without specifying the number of items is a topic of statistical debate (Fayers and Machin 2000). However, analysis of internal consistency with individual items deleted from the subscales did not consistently reveal problematic items and did not suggest any dramatic improvements. Items that were dropped to produce the IPQ-R38 (Moss-Morris et al. 2002) were not indicated as problematic in the IPQ-R50 in these samples. It would have been informative to conduct a confirmatory factor analysis on this data to test the hypothesised factor structure of the questionnaire in this population and to indicate problematic items.

However the aim of this analysis was not to *test* the psychometric structure of the questionnaire, but merely to ensure that the measure was adequate for use in further analysis. In addition, the sample size was not sufficient for this analysis given the number of questionnaire items (Ferguson and Cox 1993). It may have been possible to combine the samples in order to raise the sample size for this analysis. However the combining heterogeneous samples is considered inappropriate since factor structures in each sample may be obscured (Hair et al. 1995).

The timeline cyclical subscale showed particularly poor internal consistency in both samples and a high number of outliers in the general population suggesting the subscale was not normally distributed. There are a number of possible explanations regarding the poor performance of this scale in these samples. The timeline cyclical subscale was designed to assess perceptions of constancy in rapidly changing illnesses such as menstrual disorders. This concept may not relevant to breast cancer or the timeframe regarding cyclical beliefs may be longer in breast cancer than that assessed with the current items (eg '*The symptoms of my illness change a great deal from day to day*'). Disease specific items developed to assess beliefs about constancy of breast cancer in the longer term (eg recurrence) may capture the concept in this context. In addition, it is possible that healthy individuals have a poor understanding of the symptom experiences of breast cancer patients. Previous research has found that healthy individuals who have not directly experienced symptoms of an illness report less symptom variability than patients and that "*an understanding of this aspect of chronic illness most commonly comes from hard personal experience*" (Schiaffino and Cea 1995, pg 544). Examining the items within the timeline cyclical subscale also raises issues concerning its validity. Items in the scale appear to assess cyclical beliefs (items 3, 34); perceptions of constancy (items 7, 13, 31) and predictability (item 4) (See questionnaire in Appendix II page A-9). It is possible that these aspects do not form a cohesive dimension but reflect a range of different illness representations. The timeline cyclical subscale has also shown low reliability in recent validation work (Moss-Morris et al. 2002, see also section 6.19.1a pages 171-172). These points raise concern regarding the nature of the subscale.

The timeline acute/chronic subscale showed marginal internal consistency in both samples and was lower than that reported in patient populations. The internal consistency of the emotional representations and illness coherence subscales were high in both samples and comparable to that reported in patient populations (Moss-Morris et al. 2002). The psychometric properties of a number of the other subscales differed between the samples. It was predicted that the IPQ-R would shower higher reliability in the increased risk sample than the general population sample. This was true for the control subscales. The internal consistency of the personal control subscale in the increased risk sample was comparable to that reported in patient populations (Moss-Morris et al. 2002). Surprisingly, the identity and consequences subscales performed better in the general population sample than the increased risk sample. The identity subscale in the general population sample showed comparable levels of internal consistency to patient populations (Moss-Morris et al. 2002).

It is possible that the order of presentation of items may have affected reliability scores. Items in this study (with the exception of the identity and cause checklists) were presented in a random order. This may have lowered internal consistency compared to the work on patient populations when the items were grouped and presented according to their subscale (Moss-Morris, personal communication).

Test re-test correlation coefficients for both versions of the subscales in the follow-up sample were high. In most subscales the longer version of the scale showed higher test-retest reliability. The personal control, treatment control, illness coherence and emotional representation subscales all showed higher 3 month test-retest reliability in this sample than that found for renal patients at 3 weeks and rheumatoid arthritis patients at 6 months (Moss-Morris et al. 2002). One would expect illness perceptions of healthy individuals to be more stable than those of patients who may experience changes in their illness over time. As would be expected test-retest reliability was higher for participants who did not report any breast cancer related experiences between completing the questionnaires. This is consistent with the SRM that proposes that individual's beliefs are dynamic and may change with experience. The timeline cyclical subscale showed lower levels of test-retest reliability than other subscales and was non-significant in the group of participants with breast cancer

experiences between questionnaires. This indicates that either perceptions of prognosis of breast cancer were changed drastically by experience or that this measure is not reliable in this sample. Since women in this sample are not directly experiencing changes in symptoms it is unlikely that the type of experiences women reported between questionnaires would have such a strong effect on timeline cyclical subscale. Given the problems with internal consistency of this scale it is more likely that results reflect poor reliability.

Overall, these results suggest that the IPQ-R is applicable to assess perceptions of breast cancer in healthy women. Some of the subscales perform better than others and differences in internal consistency were found between the samples. The IPQ-R50 version appeared more reliable in both samples and will be utilised in subsequent analysis. Given the low internal consistency and test re-test reliability of the timeline cyclical subscale as well as the poor distribution of this scale it was omitted from further analysis. Care will be needed in the interpretation of the timeline acute/chronic subscale in all samples and the treatment control subscale in the general population samples due to low levels of internal consistency.

The results reported in this chapter do not provide information regarding the validity of the IPQ-R in these samples. Further research is necessary to examine the validity of assessment of illness perceptions in healthy populations. This will be discussed further in the Discussion Chapter (Chapter 11).

6.20.3 Perceptions of breast cancer held by healthy women: Descriptive statistics.

In general the mean values of the subscales suggested that healthy women believed breast cancer holds severe consequences, is long in duration and that they held strong emotional representations of the disease. Women believed in treatment control and to a lesser extent personal control of breast cancer. Women also reported a coherent understanding of the disease. Scores on these subscales ranged in both samples suggesting that women hold differing beliefs about breast cancer. Further analysis comparing the beliefs of the samples and investigating individual differences in beliefs will be reported in subsequent chapters (Chapter 8).

Women in both samples believed that there were a number of causes of breast cancer. The most important causes identified by participants in both samples included both controllable and uncontrollable factors (e.g. heredity, hormonal factors, diet and smoking). This confirms previous findings that healthy individuals hold multifactorial models of breast cancer and familial cancer (Payne 1990, Michie et al. 1996). The majority of causes noted in the open response format were items already on the causal list of the IPQ-R supporting the validity of these items in this context. This is consistent with previous research which has found patterns of attributions made for illness from fixed lists does not differ from those gleaned from open ended methods (Weinman et al. 2000, French et al., 2001). A few sporadic causes were mentioned that were extremely specific and not assessed by the IPQ-R.

6.20.4 Patterns of illness perceptions: Intercorrelations and Cluster analysis

Logical intercorrelations in the predicted direction were found amongst the subscales of the IPQ-R in both samples and reflected relationships reported in patient samples (Weinman et al. 1996). The majority of predicted associations were found in the increased risk sample except for associations between emotional representations and control perceptions, and associations between perceptions of personal control and other cognitive representations.

Exploratory analysis revealed that the number of causes participants believed to be associated with breast cancer was positively associated with perceptions of consequences and personal control in both samples. In addition, women in the general population sample who believed in more causes of breast cancer associated more symptoms with the disease and had a stronger emotional response to breast cancer. This is consistent with previous research that has shown a positive correlation between number of causal attributions for illness and distress (Turnquist et al. 1988). In addition, Weinman et al. (2000) found the number of causal attributions for spousal MI was associated with fear of another MI in the following year. Beliefs regarding the number of causes of breast cancer may therefore reflect increased anxiety concerning the disease and susceptibility to it.

Individual causal beliefs were associated with a number of belief subscales particularly consequences and personal control. As would be expected beliefs about personal control over breast cancer were positively associated with controllable causes and negatively associated with uncontrollable causes. There were a large number of intercorrelations within the causal items that suggested overlap between items. It may be possible to reduce these items into causal dimensions. Moss-Morris et al. (2002) outlined a principal components analysis of the causal items in the IPQ-R. They found that 4 factors accounted for 57% of the variance. These factors were: Psychological attributions (33%), risk factors (11%), immunity (7%), and accident/chance (6%). However, reducing causal items in this way has received criticism. Sutton et al. (submitted) criticised this approach for using pragmatic rather than theoretical rationale for reducing variables. It is argued that the items lack compatibility. The numbers of items encompassed within these subscales make the construct difficult to interpret and target with interventions. Given these criticisms and the limited sample size in the increased risk sample it was decided not to use a factor analytic approach but to explore the causal items independently.

Cluster analysis was used to assess the pattern of illness perceptions in both samples and also to provide a summary categorical variable for use in subsequent analysis. As predicted, participants in both the increased risk and general population samples were found to fall into discrete clusters based on their representations of the disease. Two clusters were identified in both samples. Participants in each cluster were differentiated by scores on all of the IPQ-R subscales. One cluster represented negative representations and the other positive representations (high scores on the control subscales and low scores on remaining subscales). These clusters are similar to those found in previous research on patients with breast cancer and Addisons disease (Buick 1997, Heijmans 1999). Causal items were not included in the analysis due to the large number of items and lack of clear hypotheses. However, exploratory analysis revealed that participants in each cluster responded differently to the causal items. It is possible that causal beliefs may be important components an individuals overall representation of disease that determine response to risk and is an area that requires further investigation.

Since cluster analysis is not based on statistical reasoning there are no tests to determine which solution best describes the data. It is possible that clusters could be further differentiated. However, given the clear 2 cluster solution represented on the dendrogram (Figure 6.2), similarity of solutions in both samples and comparison with previous published work the two cluster solution was deemed acceptable. The clusters will therefore be used as dichotomous variables in order to investigate the impact of experience of breast cancer on illness perceptions and also to address associations between patterns of beliefs and levels of distress in both samples.

CHAPTER 7

EXPERIENCE OF BREAST CANCER AND DISTRESS

7.1 AIMS AND RATIONALE

Chapter 6 outlined an evaluation of the measures used in the current research to assess experience of breast cancer in the family and illness perceptions. The measures were deemed to show sufficient reliability to continue with the analysis plan summarised in Chapter 4 (see Table 4.1, page 102). The main aim of the thesis was to test the mediation model (Figure 4.1, page 100). This model was tested in its constituent parts addressed in this and the two following chapters (Chapter 8-10). This Chapter reports on the section of work concerned with objective 2. and examines associations between experience of breast cancer in the family and level of general and cancer specific distress. The hypotheses for this section were outlined in Chapter 4 (4.1.3a; H1-H3) and will be described in more detail here. Associations between experience of breast cancer and distress were addressed at two levels. Firstly, tests were conducted to assess differences in general and cancer specific distress between samples with different experiences of breast cancer. Secondly, associations between specific experiences of breast cancer and levels of general and cancer specific distress within both the increased risk and general population samples were examined.

7.1.1 Differences in general and cancer specific distress between samples with different experiences of breast cancer: Predictions.

Literature concerning levels of distress in women with a family history of breast cancer has been inconsistent. Some studies have reported higher levels of general distress in women with a family history of the disease compared to the general population (Kash et al. 1992, Gagnon et al. 1996) whilst others have reported comparable levels of distress (eg Lerman et al. 1994a, Lloyd et al. 1996). A caveat in this research is the lack of control for possible experiences of breast cancer within the general population. For the purpose of this study therefore, women at increased risk of breast cancer were compared to a control sample of women with no experience of breast cancer in their social environment. It was hypothesised (H1) that women at increased risk of breast cancer would show higher levels of cancer specific distress because of their vulnerability to the disease and experiences in their family. Greater experience of the disease is likely to increase accessibility of thoughts about

the disease (see Chapter 3, section 3.2, page 73-74) and models of health anxiety have suggested that external events may trigger intrusive thoughts about illness (Salkovskis and Warwick 1986). In addition, Freeston et al. (1994) reported associations between subjective probability of threat occurrence with thought frequency and difficulty removing health related intrusive thoughts in a healthy population. A proportion of the general population sample were expected to show intrusive thoughts about breast cancer since health related intrusive thoughts have been reported in general population samples (Freeston et al. 1994).

7.1.2 Experience of breast cancer and level of general and cancer specific distress in the increased risk sample: Predictions.

The second level of investigation was to examine associations between specific experiences of breast cancer in the family and levels of general and cancer specific distress in the increased risk sample. Whilst the development of measures of cancer specific distress has proved useful for research in this area (See Chapter 1, section 1.4, page 48) there has been little work examining the how different types of distress develop. This analysis aimed to assess what experiences of breast cancer in the family were associated with both general and cancer specific distress. It was hypothesised (H2) that experiences of breast cancer would be associated with levels of both general and cancer specific distress. It was expected that the results would replicated findings of the pilot work (see Chapter 6, part 2, sections 6.3-6.8). Level of general distress was predicted to be associated with bereavement and diagnosis from breast cancer in the family and the recency of these events. Cancer worry was predicted to be positively associated with the degree of perceived resemblance between the respondent and their index relative and negatively correlated with how well the index relative coped with their illness and positive aspects of the experience. Both general distress and cancer worry were expected to be associated with how traumatic the experience had been and the impact of the experience on the respondent's life including family roles, relationships and life plans.

Intrusive thoughts about breast cancer were also expected to be related to different aspects of experience than cancer worry. Intrusive thoughts associated with bereavement have been considered to contribute to increased distress in women with

a family history of breast cancer (Zakowski et al. 1997). In addition, individuals experiencing multiple loss have been found to report recurrent thoughts and images regarding the experience (Nord 1996). Intrusive thoughts about breast cancer were therefore predicted to be associated with the experience of bereavement (including recency of bereavement and multiple bereavement due to breast cancer) as well as how traumatic the experience had been. Both intrusive thoughts and avoidance are characteristic markers of stress response in cancer patients and are positively correlated (Cordova et al. 1995). Measures of intrusion and avoidance are therefore often summed or considered separately. Few studies have examined differences in patterns between these constructs (Primo et al. 2000). Analysis was therefore conducted using the total score on the Impact of Event scale as an overall measure of stress response to breast cancer risk as well as separate scores on the intrusion and avoidance subscales in order to explore the associations between experience of breast cancer and these constructs.

From a clinical perspective it is important to determine aspects of experience that best predict distress. This will enable clinicians to identify vulnerable women at an early stage in the genetic counselling process and help predict potential psychological problems. Exploratory analysis was therefore conducted to determine what aspects of experience best predicted levels of general and cancer specific distress in women at increased risk of breast cancer.

7.1.3 Experience of breast cancer and level of general and cancer specific distress in the general population sample: Exploratory analysis.

A final aim of this section was to explore the impact of experiences of breast cancer on levels of general and cancer specific distress in women in the general population. Previous work has indicated that the *experience* of breast cancer in the family is associated with increased perception of risk rather than the knowledge that a family history of the disease is an established risk factor (Drossaert et al. 1996, Absetz et al. 2000). It is possible therefore that women in the general population with experiences of breast cancer would show higher levels of cancer specific distress than women without any experience of breast cancer. The impact of experiences of breast cancer in family, friends and at work on levels of general and cancer specific distress is

explored in the general population sample. It was hypothesised (H3) that experiences of breast cancer reported by women in the general population would be associated with levels of cancer specific distress.

7.2 MEASURES AND SAMPLE

Data for this analysis were derived from the cross-sectional questionnaire study. The design and procedure were previously described in Chapter 4. The samples reported in this study included: Increased risk sample (Sample A), general population sample (Sample B&C) and control sample (Sample C). These samples were described in Chapter 5.

The measures utilised in this analysis were:

- Background demographics (age, marital status, education and risk perception) (see Chapter 4. section 4.4.1, page 104)
- Experience questionnaire (increased risk sample only) (see Chapter 6, Part 2, section 6.3-6.8)
- Experience items for the general population sample (see Chapter 4, section 4.4.2b, page 105)
- General distress (GHQ-30) (see Chapter 4, section 4.4.4a, page 106)
- Cancer specific distress (Cancer worry scale, Impact of Event scale) (see Chapter 4, section 4.4.4b, page 107)

7.3 STATISTICAL METHODS

7.3.1 Univariate analysis

Chi-square test was used to compare samples on nominal data and to examine associations between categorical measures of distress and experience. T-tests were used to compare samples on continuous measures and examine associations between levels of general and cancer specific distress and experience. Pearsons correlations were also examined when both measures of experience and distress were continuous.

7.3.2 Multiple Regression

7.3.2a Background

Multiple regression is a set of techniques that examines the effects of several independent (predictor) variables on one dependent (outcome) variable. The aim of multiple regression is to derive a linear equation that relates the dependent variable with the independent variables. Multiple regression may be used for prediction and is also used to establish the comparative importance of independent variables in determining outcome variables. The independent variables are assigned different weights (regression coefficients) and the aim of analysis is to arrive at a set of regression coefficients that bring the predicted dependent values from the equation as close as possible to the observed dependent variable:

$Y = A + B_1X_1 + B_2X_2 + \ldots..B_kX_k$

Y = predicted value of the DV

A= intercept

X= Independent variables (of which there are $_K$).

B= coefficients assigned to each of the independent variables in the equation.

Adequate sample size is an important requirement of multiple regression. The sample size must be sufficient to obtain statistical power for the analysis given the number of independent variables, expected effect size and alpha level. The ratio of cases to independent variables should be high. A rule of thumb to determine the minimum sample size is a ratio of 5 observations for each independent variable, although a ratio of up to 20 is more desirable (Hair et al. 1995).

Multiple regression also has a number of assumptions. Firstly the dependent variable must be normally distributed. It is not however assumed that the independent measures are normally distributed and hence dichotomous variables can be entered as possible predictors in multiple regression models. Normality of dependent variables can be assessed by examination of skew and kurtosis values as well the graphical distribution of variables in box plots and normal probability plots. Residuals (differences between the obtained and predicted scores) must be normally distributed

about the predicted scores (normality), have a straight line relationship with predicted scores (linearity) and show equal variance about the predicted scores (homoscedasticity). These assumptions are assessed by examining normal probability plots of residuals, scatterplots of the residuals and predicted dependent variables scores and details of residual outliers (Tabachnick and Fidell 1996).

Multiple regression is also influenced by associations between independent variables particularly multicollinearity. Multicollinearity refers to the correlation amongst 3 or more independent variables. This reduces and confounds the predictive power of the variables. An initial assessment of multicollinearity involves examining the correlations amongst the independent variables to ensure that no high correlations exist ($r > .9$). Another common measure of multicollinearity is tolerance. Tolerance represents the degree to which an independent variable is explained by another independent variable. A low tolerance value denotes high collinearity. A common cut off value for tolerance is >0.1 (Hair et al. 1995).

7.3.2b Selecting independent variables in the model

Multiple regression is best conducted when each independent variable is correlated with the dependent variable but uncorrelated with other independent variables (Everitt 1996). For each independent variable the regression coefficients represent the change in dependent variable, associated with a unit change in the independent variable, conditional on other independent variables in the model remaining unchanged. It is unlikely that all variables entered into the model will contribute significantly to prediction of the dependent variable. However, all variables are conditional on other variables in the model and if one variable is removed then the regression coefficients of remaining variables will change.

There are a number of techniques available to determine which variables to include in the model. These differ in the way the variables enter the equation, what happens to variance shared by variables and how the order in which the variables are entered into the equation is determined (Tabachnick and Fidell 1996). In standard multiple regression all independent variables are entered into the regression equation at once and each assessed as if all other variables have already been entered. Hierarchical

regression requires that the researcher specify the order in which variables are entered into the model. Variables are examined in terms of what each adds to the equation at that point of entry. Decisions about the order of variables should be based on sound logical or theoretical arguments. Statistical regression is a set of methods in which the variables are entered depending on statistical criteria. There are three statistical regression methods available: forward regression, backward regression and stepwise regression. These are outlined as follows.

- Forward selection: Variables are entered one at a time if they significantly contribute to the equation.
- Backward selection: The equation starts with all variables in and deletes variables if they do not contribute to the equation.
- Stepwise: The equation starts out empty and variables are entered according to forward selection. At each stage all variables may also be considered for removal using backward selection. It is possible that a variable included earlier in the model may be removed at a later stage if its contribution to the model is no longer significant following the entry of additional variables.

Of these approaches the Stepwise method has been considered the best approach to obtaining the optimal prediction solution (Tabachnick and Fidell 1996). When using multiple regression in order to test causal mechanisms and in search for explanation the standard multiple regression technique is most appropriate.

7.3.2c Predicting level of distress from experience variables

Multiple regression analysis of experience variables was used to determine the best predictors of psychological well-being in the increased risk sample. The analysis was conducted in the following steps:

Step 1.

Experience items that were correlated with the dependent variable ($p<0.1$) were selected for inclusion in the model. Both continuous and binary variables were considered for inclusion. A number of variables (e.g. recency of bereavement of index relative) were only available for individuals who had particular scores on a binary variable (eg is the index relative alive or dead). This raised problems of

entering this variable in the analysis while also maintaining the full sample size. In these circumstances a missing data dichotomy was used (Cohen and Cohen 1983). Two variables are entered into the model. One binary variable represents the missing data dichotomy (scores 1 for missing data (eg index relative is alive) and 0 for participant with data (eg index relative has died)). The second variable is the continuous variable (eg recency of bereavement). Individuals without scores on this variable are provided with an arbitrary constant instead of missing values and hence remain in the analysis. This analysis can be conducted using stepwise regression if the missing values are replaced with the mean value (Cohen and Cohen 1983). Both hierarchical analysis and stepwise regression were conducted. The results of these analyses were comparable and the stepwise regression are reported.

Step 2.
Checks were made to ensure the sample size was sufficient for the analysis. During study design a sample size of 100 in both the increased risk and control sample was anticipated in order to obtain sufficient power for multiple regression. A sample size of 100 allows a low R-squared value (.10) to be deemed as statistically significant with a power of .80 and the significance value set at 0.05 (Hair et al. 1995). A sufficient number of cases were available to predict GHQ and cancer worry scores. However only 61 participants fully completed the Impact of Event scale and sample size was likely to be further reduced by missing data on independent variables.

Step 3.
Normality of dependent variables was investigated by assessing skew and kurtosis. The GHQ scale was skewed and hence multiple regression was conducted both on the raw scores and also on a logarithmic transformation of scores. The results of the analyses were comparable and hence results from the raw score are reported for simplicity. The cancer worry scale showed sufficient normality for this analysis.

Step 4.
Correlations were examined amongst the independent variables to check for multicollinearity to ensure that no variables were extremely highly correlated ($r > .9$). Further to this the tolerance was examined in the output using cut off at 0.1 (Hair et al. 1995).

Step 5.
Stepwise multiple regression analysis was conducted since the main aim of the analyses was to determine the best predictor(s) of psychological well-being.

Step 6.
Residual scatterplots and normal probability plots were examined for multivariate normality (Normality, linearity, homoscedasticity and independence of residuals). Details of residual outliers were also examined. These assumptions were met for the multiple regression equations predicting GHQ and cancer worry. Multivariate normality for the Impact of Event analyses was problematic. Scatterplots suggested that the residuals were not normally distributed around the dependent variable in this model and the normal probability plot of the residuals from the intrusion subscale suggested that the residuals were not normally distributed. Given these problems and reduced sample size, it was decided that the Impact of Event scale did not meet the requirements for multiple regression.

Step 7.
The statistics to be reported from this analysis were:

- Significance of model ($p<0.05$).
- Adjusted R Square (% of variance of the dependent variable accounted for by the model, adjusted to account for the number of predictor variables in the model).
- Significance of variables in the model ($p<0.05$).
- Final equation standardized beta coefficients. (Standardized regression coefficients representing change in the standardized dependent variable produced by a change of 1 SD in the independent variable. Allows for direct comparison of the relative explanatory power of predictor variables.)

7.4 RESULTS

7.4.1 Descriptive statistics

Background demographic data for each sample were provided in Chapter 5 (see section 5.3, page 118). Table 7.1 shows the distribution of scores on distress measures for each sample.

Table 7.1- Distress scores in the increased risk (A) and general population sample (B&C).

Measure	Descriptive statistic	Increased risk sample (A)	General population sample (B&C)
GHQ	n	116	253
	Mean (sd)	27.0 (11.86)	27.9 (12.20)
	Range	9-70	5-76
Cancer worry	n	116	253
	Mean (sd)	10.7 (2.58)	9.1 (2.27)
	Range	6-19	6-18
Impact of Event- Total score	n	58	65
	Mean (sd)	18.2 (12.69)	14.8 (15.15)
	Range	0-54	0-59
Impact of Event- Intrusion subscale	n	60	68
	Mean (sd)	8.1 (5.94)	5.9 (5.97)
	Range	0-25	0-23
Impact of Event- Avoidance subscale	n	58	65
	Mean (sd)	10.3 (8.47)	8.8 (9.68)
	Range	0-34	0-23

7.4.2 Distress and background demographics

7.4.2a Increased risk sample (A)

Age was not significantly correlated with GHQ score or scores on the Impact of Event scale. There was a trend for age to be negatively correlated with cancer worry score in this sample ($r = -.17$, $n = 116$, $p = 0.068$). Marital status and education were not associated with any distress measure. Risk perception as measured on a 5 point Likert scale (see section 4.4.1, page 105) was significantly correlated with GHQ

score (r= .18, n= 115, p= 0.049) and cancer worry (r= .43, n= 115, p<0.001). Participants who reported to have thought about breast cancer in the previous week reported higher personal perception of risk (mean= 3.1, sd= 0.74, n= 60) than those who did not (mean= 2.8, sd= 0.69, n= 56). However there were no significant correlations between scores on the Impact of Event subscales and risk perception (p<0.05).

7.4.2b General population sample (B&C)

Age, marital status and education were not associated with any of the distress measures. Risk perception as measured on a 5 point Likert scale (see section 4.4.1, page 105) was significantly correlated with cancer worry score (r= .29, n= 249, p<0.001). Participants who reported to have thought about breast cancer in the previous week reported higher personal perception of risk (mean= 2.6, sd= 0.74, n= 68) than those who did not (mean= 2.3, sd= 0.59, n= 181). Both the intrusion and avoidance subscales were positively correlated with risk perception (p<0.01).

7.4.3 Comparing levels of distress in samples with different experiences of breast cancer.

7.4.3a General distress

A GHQ 'case' is a score >5 on the GHQ-30. Thirty-six (30.8%) of participants at increased risk of breast cancer (Sample A) and 31 (31%) of the control sample (Sample C) without any experience of breast cancer scored >5 on the GHQ. There was no significant difference between the control sample and the increased risk sample in the proportion of GHQ 'cases' (chi-square = .009, df= 1, p= 0.93).

There was no significant difference in mean GHQ scores between the increased risk sample (Sample A) (n= 116, mean= 27.0, sd =11.86) and the control sample (Sample B) (n= 98 , mean= 27.4, sd= 10.72); (t=.24, df= 212, p= 0.81).

7.4.3b Cancer specific distress

7.4.3b(i) Cancer worry

Women in the increased risk sample scored significantly higher on the cancer worry scale (n= 116, mean= 10.7, sd= 2.58) than women in the control sample (n= 99, mean= 8.9, sd= 2.0); (t= 5.56, df= 213, p< 0.001).

7.4.3b(ii) Impact of Event scale

More women in the increased risk sample had thought about breast cancer in the past week (52.1%) than in the control sample (23%) (chi-squared= 18.85, df= 1, p<0.001).

The mean score for each sample on each of the subscales are provided in Table 7.2. There was a trend that suggested that women in the increased risk sample (A) had higher intrusion scores than women in the control sample (C) (t= 1.93, df= 81, p= 0.057). Although the increased risk sample also showed higher total scores and avoidance scores these differences were not significant (p<0.05).

Table 7.2- Mean scores for each sample on the Impact of Event scale.

Measure		**Increased risk sample (A)**	**Control sample (C)**
Total score	n Mean (sd)	58 18.2 (12.69)	21 14.1 (12.8)
Intrusion subscale	n Mean (sd)	60 8.1 (5.94)	21 5.4 (4.74)
Avoidance subscale	n Mean (sd)	58 10.3 (8.47)	23 8.3 (8.68)

7.4.4 Experience of breast cancer and levels of distress in the increased risk sample (A)

7.4.4a General distress

Differences between GHQ 'cases' and 'non-cases' are summarised in Table 7.3. GHQ 'cases' were likely to have lost their index relative from breast cancer

significantly more recently than 'non-cases' ($p<0.01$). They were also more likely to have suffered bereavement in their family from breast cancer significantly more recently than 'non-cases' ($p<0.05$). GHQ 'cases' reported that they personally knew significantly more relatives who had suffered from breast cancer than non-cases ($p<0.05$). There was also a trend to suggest that 'cases' felt their life plans had changed more because of the risk of cancer in their family compared to 'non-cases' ($p=0.055$).

Table 7.3- Differences in experience between GHQ 'cases' and 'non-cases' in the increased risk sample (A).

Experience item		**GHQ 'case'**	**'Non case'**	**df**	**t**	**p**
Recency of bereavement of index relative from breast cancer (if any) (months)	N Mean SD	22 124.5 107.12	48 219.02 136.88	68	2.86	.006
Number of relatives personally known who had suffered from breast cancer	N Mean SD	36 2.5 1.34	80 1.9 1.17	114	2.39	.019
Most recent bereavement from breast cancer (if any) (years)	N Mean SD	25 11.6 11.42	59 17.9 11.95	83	2.44	.017
'How much do you feel your life plans have changed because of the risk of cancer in your family'?	N Mean SD	36 2.1 1.18	78 1.7 0.96	112	1.94	.055

The effect of recency of bereavement on general distress was confirmed in correlational analysis. GHQ score was significantly negatively correlated with recency of bereavement of a relative from breast cancer ($n= 85$, $r=-.31$, $p= 0.004$). For participants whose index relative had died from breast cancer GHQ score was correlated with recency of bereavement of index relative ($n= 71$, $r=-.26$, $p= 0.032$). A few trends were identified in the data. A negative correlation between GHQ score and experience item '*To what extent have your experiences been positive*?' ($n=114$, $r=-.18$, $p= 0.059$) was identified. There was also a trend for participants whose index relative was their mother to score lower on the GHQ ($n= 80$, mean = 25.8, sd =

11.67) than participants whose index relative was not their mother (n= 36, mean= 29.9, sd= 11.96); (t= 1.74, df= 114, p= 0.084).

7.4.4b Cancer specific distress

7.4.4b(i) Cancer worry

Individuals who scored higher on the cancer worry scale were more likely to report that their relatives illness had been traumatic (n=109, r= .22, p= 0.021) and that their life plans (n= 114, r= .36, p< .001) and role in the family (n= 114, r= .21, p= 0.027) had been changed because of the risk of cancer in the family. Participants who reported higher levels of cancer worry were also likely to have been younger when their index relative was diagnosed (n= 116, r= -.19, p= 0.044).

7.4.4b(ii) Impact of Event scale

Table 7.4 shows differences in experience between participants who did and did not report to have thought about breast cancer in the previous week. Individuals who reported to have thought about breast cancer in the previous week and completed the Impact of Event scale reported significantly more changes in their life plans because of their experiences ($p<0.05$). There was also a trend for this group to have been younger when their relative was diagnosed, to report fewer positive aspects about the experience and also to report that their role in the family had changed ($p<0.1$).

Table 7.4- Differences in experience between participants in the increased risk sample who did and did not complete the Impact of Event scale.

Experience item	Thought about breast cancer in the previous week		Did not think about breast cancer in the previous week	df	t	p
'How much do you feel your life plans have changed because of the risk of cancer in your family'?	n Mean sd	59 2.0 1.07	56 1.6 1.02	113	2.005	.047
Participants age when index relative was diagnosed	n Mean sd	61 19.9 10.96	56 23.6 12.35	115	1.71	.090
'To what extent have your experiences been positive'?	n Mean sd	59 2.3 1.03	56 2.6 1.20	113	1.70	.091
'Do you feel that your role in the family has changed because of your experiences of breast cancer'?	N Mean SD	59 2.6 1.32	56 2.2 1.44	113	1.667	.098

Correlational analysis revealed that individuals with higher scores on the Impact of Event scale were more likely to report that their life plans had changed because of the risk of cancer in their family ($n = 114$, $r = .36$, $p < 0.001$). There was also a trend for individuals with higher scores to report that they were closer to their index relative ($p<0.1$) and that they found the experience more traumatic ($p<0.1$).

Individuals scoring higher on the intrusion subscale were more likely to report that their life plans had changed because of the risk of cancer in their family ($n = 58$, $r.= 40$, $p = 0.002$) and that they resembled their index relative ($n = 57$, $r = .26$, $p = 0.049$). There was also a trend for participants with higher intrusion scores to report that they were closer to their index relative ($p<0.1$). There was a trend for individuals with higher scores on the avoidance subscale to report they found the experience more traumatic ($p<0.1$).

7.4.5 Predicting distress in the increased risk sample (A) from experience items

7.4.5a Predicting GHQ score from experience items.[7]

A stepwise multiple regression analysis was used to predict GHQ score from the experience variables that were associated with it ($p<0.1$). The items that were considered for entry into the model were:

- Recency of bereavement of a personally known relative from breast cancer (plus dichotomous variable categorising if participants had or had not experienced a recent bereavement) (see 7.3.2c, step 1, page 204).
- Recency of index relative bereavement (plus dichotomous variable categorising if participants had lost their index relative from breast cancer or not)
- If the index relative was the participant's mother or another relative (dichotomous variable)
- Subjective experience item 12: '*To what extent have your experiences been positive?*'

The recency of bereavement variables showed a strong correlation ($n= 71$, $r= .92$, $p< 0.001$), which would lead to problems of multicollinearity in a multiple regression analysis (see 7.3.2a, page 203). It was decided to choose one of these variables for use in the model. More participants provided data concerning the recency of bereavement of any relative they personally knew (n=86) than the recency of bereavement of the index relative (n=78). Models were compared using both variables and the recency of bereavement of any relative personally known to the respondent accounted for more variance in GHQ score suggesting this was the better variable to use.

The results of the regression model are provided in Table 7.5. The results indicated that two of the experience variables were significant predictors of GHQ score:

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[7] A similar model was obtained using a logarithmic transformation of GHQ score

Recency of bereavement of a personally known relative from breast cancer and the extent to which participants reported their experience had been positive ($p<0.05$). Both of these items showed negative beta coefficients indicating that having experienced bereavement more recently and not perceiving the experience to have been positive predicted higher GHQ scores. Together these variables accounted for 9.9% of the variance in GHQ score ($p<0.01$).

Table 7.5- Stepwise regression predicting GHQ score from experience items in the increased risk sample.

Adj R square	**F**	**p**	**Significant variables**	**Std beta**	**t**	**p**
.099	7.01	.001	Recency of bereavement of a personally known relative from breast cancer.	-.30	-3.26	.001
			Item 12 *'To what extent have your experiences been positive?'*	-.19	-2.11	.037

7.4.5b Predicting cancer worry score from experience items

A stepwise multiple regression analysis was used to predict cancer worry score from the experience variables that were associated with it ($p<0.1$). The items entered into the model were:

- Participant's age when their index relative was diagnosed with breast cancer.
- Subjective experience item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'*
- Subjective experience item 9: *'Do you feel that your role in the family has changed because of your experiences of breast cancer?'*
- Subjective experience subscale: Traumatic experience.

The results of the regression model are provided in Table 7.6.[8] The results indicate that one experience item was a significant predictor of cancer worry score: Subjective experience item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'* The positive beta coefficient suggested that higher cancer worry scores were predicted by higher scores on the experience item. This item accounted from 12.3% of the variance in cancer worry scores.

Table 7.6 Stepwise regression predicting cancer worry score from experience items in the increased risk sample.

Adj R square	F	p	Significant variables	Std beta	t	p
.123	16.10	.000	Item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'*	.36	4.01	<0.001

7.4.6 Experience of breast cancer and levels of distress in the general population sample (B&C)

7.4.6a Experience of breast cancer in the general population sample.

One hundred and fifty-six (60.9%) respondents in the general population sample reported some experience of breast cancer (Sample B). Sixty-four (25%) reported that a relative had suffered from breast cancer. Fourteen of these participants reported that a first degree relative had suffered from the disease and 20 reported that more than one relative had suffered from breast cancer. Fifty-one respondents (19.9%) reported that a family member or close friend had suffered from breast cancer recently. Thirty-one participants (12.1%) reported experience of breast cancer at work and ninety (35.2%) reported 'other' experiences of breast cancer.

[8] Age was correlated with cancer worry ($p<0.1$) (see 7.4.2a, page 207). When age was included as a predictor the model was unchanged.

7.4.6b General distress

There was no difference in the proportion of GHQ 'cases' between participants who reported any breast cancer experience (Sample B) (n= 156, GHQ 'cases' = 34.8%) and the control sample (Sample C) (n= 100, GHQ 'cases' = 31.6%); (chi-square = .28, df= 1, p= 0.60). There was no significant difference on GHQ score between participants who reported any experience of breast cancer (Sample B) (n= 155, mean= 28.3, sd= 13.1) and the control sub-sample who reported no experiences of breast cancer at all (Sample C) (n= 98, mean= 27.4, sd = 10.7); (t= .57, df= 251, p= 0.572). Specific experiences of breast cancer in this sample were not associated with general distress ($p<0.05$).

7.4.6c Cancer specific distress

7.4.6c(i) Cancer worry

There was no difference in cancer worry between participants who reported any experience of breast cancer (Sample B) (n= 159, mean= 9.2, sd= 2.42) and the control sub-sample (Sample C) (n= 99, mean= 8.9, sd= 2.01); (t= 1.22, df= 251, p= 0.221). However specific types of experience were associated with levels of cancer worry (see Table 7.6). Participants who reported that a family member or close friend had suffered from breast cancer recently reported higher levels of cancer worry than those who did not report a recent experience of breast cancer ($p<0.05$). Participants who reported a recent experience of breast cancer also reported higher levels of cancer worry than the control sample without any experience of breast cancer (n= 99, mean= 8.9, sd = 2.01); (t= 2.11, df= 147, p= 0.017). Participants who reported that a first degree relative had suffered from breast cancer showed a trend to score higher on the cancer worry scale (n= 14, mean= 10.2, sd= 2.42) than participants whose relative who had suffered from breast cancer was not a first degree relative (n= 44, mean= 8.6, sd= 2.11); (t= 1.94, df= 56, p= 0.057).

7.4.6.c(ii) Impact of Event scale

There were no differences in the proportion of individuals who reported to have thought about breast cancer in the previous week between the sample who reported *any* experience of breast cancer (Sample B) (30.5%) and the control sub-sample (Sample C)(23.2%) (chi-square= 1.60, df= 1, p= 0.21). However, 48% of those who

reported a family member or close friend had suffered from breast cancer *recently* reported to have thought about breast cancer in the previous week, compared to 22.8% of those who reported not to have a family member or close friend who had suffered from breast cancer recently (chi-square= 12.71, df=1, p< 0.001). Those who reported a recent experience were also more likely to have completed the Impact of Event scale compared to those without any experience of breast cancer at all (23% completed the scale); (chi-square= 9.44, df= 1, p= 0.002). Just under ten percent (9.7%) of those who reported to have contact with cancer patients at work reported to have thought about breast cancer in the past week compared to 30.5% of those who reported no experience of cancer at work (Chi-square= 5.83, df= 1, p= 0.016). Over half (57.1%) of participants with a first degree relative who had suffered from breast cancer reported to have thought about breast cancer in the past week compared to 31.8% of participants who had a relative who had suffered from breast cancer who was not a first degree relative (Chi-square= 2.89, df= 1, p= 0.09).

Participants who reported that a family member or close friend had suffered from breast cancer recently reported higher levels of intrusive thoughts about breast cancer ($p<0.05$) and also showed a trend to report more avoidance of breast cancer ($p<0.1$) than the rest of the sample who did not report a recent breast cancer experience (see Table 7.7). Although participants who reported a recent experience of breast cancer consistently scored higher on the Impact of Event scales than the control sample without any experience of breast cancer these differences were not statistically significant ($p<0.05$). Participants who reported that more than one of their relatives had suffered from breast cancer showed a trend for higher intrusion scores (n= 9, mean= 8.8, sd =2.75) than participants who reported that one relative had suffered from breast cancer (n=10 mean= 3.90, sd= 4.07); (t= 1.82, df= 17, p= 0.09).

Table 7.7- Differences in psychological well-being in the general population samples between participants with and without a recent breast cancer experience.

Psychological well-being measure		Recent experience of breast cancer	No recent experience of breast cancer	df	t	p
Cancer worry	n	50	202	250	2.45	0.015
	Mean	9.8	8.9			
	sd	2.36	2.19			
Impact of Event-Intrusion score	n	24	44	66	2.19	0.032
	Mean	8.0	4.7			
	sd	7.42	4.73			
Impact of Event-Total score	n	23	42	63	2.05	0.044
	Mean	19.8	12.0			
	sd	18.66	12.19			
Impact of Event-Avoidance score	n	23	42	63	1.88	0.064
	Mean	11.8	7.1			
	sd	11.58	8.16			

7.5 SUMMARY AND DISCUSSION

The results reported in this Chapter replicated some of the findings reported in the pilot work reported in Chapter 6 and confirmed a number of hypothesised relations between experience of breast cancer and levels of general and cancer specific distress at a number of levels (see section 4.1.3a, H1-H3, page 101). Firstly, the data showed differences in levels of cancer specific distress between women at increased risk of breast cancer (Sample A) compared to women in the general population with no experience of the disease at all (Sample C). Secondly, a number of associations between aspects of the experience and level of cancer specific of distress were found in both samples.

7.5.1 Differences in general and cancer specific distress between samples with different experiences of breast cancer.

The samples showed comparable levels of general distress and no differences were found in the proportion of GHQ 'cases' or scores on the GHQ between the increased

risk (Sample A) and the control sample of women with no experience of breast cancer (Sample C). As predicted, women in the increased risk sample showed significantly higher levels of cancer specific distress than the control sample. Women at increased risk of breast cancer showed higher levels of cancer worry than controls. Although a proportion of women in the control sample reported they had thought about breast cancer in the previous week, women at increased risk of the disease were more likely to have thought about breast cancer and showed higher frequency of intrusive thoughts about breast cancer as assessed by the Impact of Event scale. This replicates previous research that has identified higher levels of cancer specific distress in women with a family history of breast cancer (Zakowski et al. 1997, Lloyd et al. 1996) and confirms the prediction that women with greater experience of breast cancer are likely to experience higher levels of intrusion about the disease.

The mean cancer worry score in the increased risk sample (10.7, sd= 2.58) was slightly lower than that reported in the pilot work which assessed women prior to genetic counselling (mean = 11.2, sd= 2.81) (see Chapter 6, section 6.7.2b, page 139). The cancer worry scores in this study were also lower than those reported in other studies assessing women with a family history of breast cancer prior to genetic counselling (Watson et al. 1998, Brain et al. 1999). This may suggest that women who have received genetic counselling show lower levels of cancer worry than women who have yet to learn their risk status. This has methodological implications For example, recruiting participants at cancer clinics may inflate cancer worry scores due to anxieties about the appointment. No published information from this scale in the general population is available for comparison to date.

The opt out box in the Impact of Event scale allowed respondents to omit completing the scale if they had not thought about breast cancer in the past week (Lloyd et al. 1996). This proved useful since nearly 50% of the increased risk sample and over 70% of the general population sample reported they had not thought about breast cancer in the past week. Although this supported suggestions that this strategy should be utilised (Thewes et al. 2001) omitting data on these participants dramatically reduced the sample size associated with this scale making further multivariate analysis problematic. Comparison to published data proves difficult since mean

scores are inflated when including only those who had thought about breast cancer in the past week. Other studies that do not include the opt out box allow individuals who do not report any intrusive thoughts or avoidance of breast cancer to score zero on the weighted scale. Assigning a score of zero for those who reported not to have thought about breast cancer in the past week allowed comparison with other studies that have used this measure. These comparisons indicated that levels of intrusive thoughts and avoidance of breast cancer as assessed by the Impact of Event scale in the increased risk sample were comparable to those reported by women in a 3 month follow up after breast cancer risk counselling (Lerman et al. 1996). Scores were lower than those reported by women prior to attending for genetic counselling (Watson et al. 1999) and also by women attending the Western General Hospital one stop symptomatic breast clinic (Leithead 2000). Scores were higher than those reported by the general population with no trauma history (Briere and Elliott 1998). Although these comparative values are logical it was decided not to use data from the whole sample in future analysis because the scores were extremely positively skewed towards zero. Future studies using this measure with the opt out box would need to estimate the proportion of the sample likely to complete the scale and calculate the sample size required for statistical analysis accordingly.

7.5.2 Experience of breast cancer and levels of general and cancer specific distress in the increased risk sample.

As expected measures of general and cancer specific distress were associated with different experiences of breast cancer in the family. Case level distress as assessed by the GHQ was associated with recency of bereavement. This replicates the results from the previous pilot work (Chapter 6, section 6.7.4a, page 139-140) in which *recency* of bereavement rather than occurrence of bereavement was associated with levels of general distress. However no associations were found between general distress and the experience of bereavement or diagnosis in the family, maternal bereavement or multiple bereavement. General distress was associated with recency of bereavement of both an affected relative and the index relative. High correlations between these variables however, suggested that many respondents had referred to the same event. General distress was also associated with the number of personally known relatives to have suffered from breast cancer an effect that was not identified

in the pilot study. This may be due to differences in the samples. In this study women knew more relatives who had suffered from breast cancer and also were aware their family history was a significant risk factor.

General distress was found to be negatively associated with the extent to which the participant regarded their experience as having been positive. Research on cancer patients has shown that those patients who are able to focus on positive aspects of the experience show reduced emotional distress and this may reflect a coping mechanism for dealing with threatening events (Dunkelschetter et al. 1992, Taylor and Armor 1996). Women at increased risk of breast cancer who are unable to utilise this coping mechanism may be more prone to increased levels of general anxiety. In addition clinical work has shown that negative feelings and dissatisfaction with caregiving may result in protracted bereavement reactions and heightened distress following bereavement (Kurtz et al. 1997). It is not possible to determine if this association reflects negative feelings regarding the experience or the ability to focus on positive aspects of the experience as a coping mechanism.

The second unexpected association revealed in the analysis was that participants whose index relative was not their mother showed higher levels of general distress. This result was surprising since participants who chose their mother as their index relative reported to be closer to them than did participants who chose another relative as their index relative ($p< 0.001$) and is inconsistent with previous research that has indicated the distressing nature of maternal breast cancer (Wellisch et al. 1991, 1992, 1996, Julian-Reynier et al. 1999, Erblich et al. 2000). The finding may have reflected recency of bereavement of the index relative. In order to complete the questions individuals are likely to have chosen a recent experience they can remember well. Those who chose a relative other than their mother were significantly more likely to have lost this relative more recently (mean months= 85.6) compared to those who chose their mother (mean months= 220.9); ($t= 4.07$, $df= 76$, $p< 0.001$). Given the strong effect of recency of bereavement on levels of general distress it is possible that the effect of the relation of the index relative is a function of recency of the experience. It may also be the case that losing a relative from the same generation

(e.g. sister) may provoke stronger issues about ones own mortality than losing ones mother.

Multiple regression analysis revealed that general distress was best predicted by recency of bereavement and the extent to which the respondent believed the experience to be positive. Together these variables only accounted for a small portion of the variance (9.9%). However, given the vast number of factors in other areas of life that contribute to general distress this effect is notable.

A number of predicted associations were found between cancer specific distress measures and aspects of experience. As expected cancer worry was found to be significantly associated with how traumatic the respondent reported their index relative's experience to have been and the degree to which their life plans and role in the family had been changed. Women who were younger when their index relative was diagnosed also showed higher levels of cancer worry. This effect has been inconsistent in the literature (Wellisch er al 1992, Erblich et al. 2000) and was not identified in the pilot work (Chapter 6). As in the pilot work (Chapter 6, section 6.7.4b, page 140-141) no association was found between cancer worry and perceived resemblance with the index relative although women who perceived themselves as more similar to their index relative, did show more frequent intrusive thoughts about breast cancer as assessed by the Impact of Event scale. Resemblance to relatives may serve as a reminder of personal risk status and therefore increase accessibility of thoughts concerning the disease although it was interesting that this did not translate into cancer worry as predicted. A greater understanding of issues related to perceived resemblance needs to be examined in more depth in order to understand these associations. For example a number of moderating factors may be critical. The association between perceived resemblance and distress may be moderated by a number of factors including beliefs about inheritance and genetics. The predicted association between cancer worry and positive aspects of the experience was not identified. Although positive interpretation of the experience may be an effective coping strategy for reducing distress about a traumatic event it may not reduce concern about ones own risk.

Cancer worry was best predicted by the degree to which participants felt their life plans had changed because of the risk of breast cancer in their family. This accounted for 12% of the variance in cancer worry score. This item was also associated with intrusive thoughts about breast cancer as assessed by the Impact of Event scale. Although this item was designed to reflect past changes in life plans because of breast cancer experiences following work by Wellisch et al. (1991, 1992) it is possible that this item also captured women's concern about future changes in life plans. Many women in this study had young families and may be concerned for the future of their family if they were to develop breast cancer. A qualitative study addressing how women cope with living breast cancer risk identified high levels of concern over the future of their family if they were to develop the disease (Appleton et al. 2000). The fairly high proportion of variance in cancer worry accounted for by this one item may reflect concern over future life events as well as past breast cancer related experiences.

Only a few experience items were associated with intrusive thoughts about breast cancer assessed by the Impact of Event scale. This was surprising since experiences were considered to prompt images of breast cancer and intrusive thoughts about the disease. No associations were found between intrusive thoughts and experience of bereavement as has been reported in other studies (Zakowski et al. 1997, Erblich et al. 2000). The majority of questions in the experience questionnaire were concerning experiences in the past. Although these experiences were often related to levels of cancer worry it is possible that more recent or current experiences (e.g. BSE, attending the clinic) would promote intrusive thoughts. No experience items were significantly associated with the avoidance subscale of the Impact of Event measure. It is therefore unclear what factors contribute to avoidance in this sample.

7.5.3 Experience of breast cancer and levels of general and cancer specific distress in the general population sample.

As hypothesised (H3), specific experiences of breast cancer in the general population sample were also associated with levels of cancer specific distress. A recent experience of breast cancer in family or friends was found to be associated with both cancer worry and intrusive thoughts about breast cancer. Women who had a recent

experience in the general population were as likely to have thought about breast cancer in the previous week as the increased risk sample (57%). This confirms previous studies that have indicated that external stimuli can act as cues which trigger intrusive health related thoughts (Freeston et al. 1994). However not all experience have the same effect. Experience of breast cancer at work in this sample was associated with reduced cancer specific distress. This may reflect habituation with exposure to breast cancer. As in the increased risk sample no significant associations were found between avoidance and experience of breast cancer in the general population.

CHAPTER 8

EXPERIENCE OF BREAST CANCER AND ILLNESS PERCEPTIONS

EXPERIENCE OF BREAST CANCER AND ILLNESS PERCEPTIONS

8.1 AIMS AND RATIONALE

The previous Chapter reported associations between experience of breast cancer in the family and levels of distress. This provided preliminary evidence for one section of the mediation model (Figure 4.1, page 100). The results reported in this Chapter refer to objective 3. and examines the section of the model linking experience of breast cancer and illness perceptions (see 4.1.2, page 99-100). The SRM proposes that a healthy individual's perceptions of disease are derived from experience with the illness within the social environment and that illness perceptions may influence response to health threats (Leventhal et al. 1980). However, few studies have investigated the origins and development of illness representations in healthy individuals.

Leedham and Meyerowitz (1999) provided evidence that experience of cancer in the family may influence perceptions of the disease. They conducted interviews with 45 women whose parents had suffered from cancer in their childhood. Interviewees reported that the experience continued to affect them in adult life, particularly their views about cancer. Nearly half of the sample reported that they had learnt how 'horrible' cancer was and 40% reported to have gained a better understanding of cancer. A quarter of respondents also reported that they appreciated the serious nature of the disease. Although it is likely that women with a family history of breast cancer have developed illness representations based on their experiences no research to date has examined illness perceptions in this population.

A summary of the aims and hypotheses (H4-H6) for this set of analyses were outlined in Chapter 4 (4.1.3b, page 101). As in Chapter 7, two levels of enquiry were adopted. Firstly, differences in illness perceptions between the increased risk (Sample A) and control sample (Sample C) were assessed. Secondly, detailed analysis of associations between experiences of breast cancer and illness perceptions in both the increased risk sample and general population sample (Sample B&C) were examined.

8.1.1 Differences in illness perceptions between samples with different experiences of breast cancer: Predictions.

Comparisons were made between the increased risk sample (Sample A) and the control sample (Sample C) who had no personal experience of breast cancer in their social environment. Firstly, differences between the sample in patterns of illness representations as indicated by the cluster solutions are explored followed by comparisons of scores on the IPQ-R dimensions and causal items.

Although there has been a lack of work addressing illness representations in healthy individuals it is possible to make predictions concerning associations between experience of breast cancer and illness perceptions based on theory. The SRM suggests that individuals who perceive themselves as vulnerable to a health threat are more likely to develop representations of the disease in question. It was therefore hypothesised (H4) that women at increased risk of breast cancer would hold stronger and more elaborate beliefs about breast cancer (ie women at increased risk will score higher on the identity, timeline acute/chronic, consequences and emotional representations subscales) than women in the control sample due to their risk status as well as greater experience of the disease. It was also predicted that women in the increased risk sample would have a better understanding of the disease due to greater access to information about breast cancer and their experiences (Leedham and Meyerowitz 1999). This would be reflected by lower scores on the illness coherence subscale compared to the control sample. Although women at increased risk were expected to show stronger beliefs about control of breast cancer it was difficult to determine the direction of this effect (ie would women at increased risk perceive greater or less control than women without experience of the disease?). This would most likely be dependant on the nature of women's experience of the disease in their family.

Previous research has shown that women in the general population perceive breast cancer as a very serious illness and overestimate the distressing nature of the condition (Roberts et al. 1984, Payne 1990, Buick 1997). Relative to this, women at increased risk might show defensive response to their risk. A defensive response to a health threat is characterised by a lowered perception of the seriousness of the health

threat by those at risk compared to those not at risk. This effect has been demonstrated in both hypothetical and real life health screening situations (Jemmott et al. 1986, Croyle et al. 1997, Rimes et al. 1999). This bias is known as 'threat minimization' and is found to occur in uncontrollable situations as a means of reducing emotional distress association with the threat (Croyle et al. 1997). It is unknown to date if individuals at increased risk of disease due to genetic factors who have close experience of illness show threat minimization.

Predictions concerning differences between the samples on causal beliefs about breast cancer were also made. Individuals who have suffered a disease themselves or are close to patients have been shown to make causal attributions for the illness (Turnquist et al. 1988, Taylor et al. 1984). It is likely that women in the increased risk sample would have searched for explanations for the occurrence of breast cancer in their family. In addition, women in the increased risk sample have attended genetic counselling and have been provided with information concerning the potential causes of breast cancer from qualified clinicians. In contrast, women in the general population have previously been found to hold misconceptions about causes of breast cancer (Payne 1990). It was predicted therefore that women in the increased risk sample would believe in a greater number of causal factors and would be more likely to believe in medically accepted risk factors (eg heredity, age, hormonal factors) (see 1.1.3, page 24) than women in the control sample.

Difference in beliefs regarding controllable and uncontrollable causes of breast cancer were also explored. Previous research has warned that genetic testing may result in fatalism (Senior et al. 1999, 2000). Although the women in the increased risk sample in this study had not received genetic testing they had received genetic counselling in which they had been informed of their increased risk of breast cancer due to a potential genetic predisposition. This may enhance deterministic beliefs about developing breast cancer. Research has also indicated that patients often make more attributions to uncontrollable factors than healthy individuals in order to avoid responsibility and negative self evaluation (French et al. 2001). Turnquist et al. (1988) found that husbands of patients with breast or cervical cancer often attribute the disease to aspects of their wives personality (Turnquist et al. 1988). Turnquist et

al. (1998) interpreted this as a protective bias to reduce responsibility for potential prevention of the disease. It was possible therefore that women in the increased risk sample may show less conviction in controllable causes of breast cancer due to fatalism or as a means to protect self esteem.

8.1.2 Illness perceptions and experience of breast cancer in the increased risk sample: Predictions.

The second aim of this section of analysis was to investigate associations between experience of breast cancer in the family and illness perceptions in the increased risk sample. Bishop and Converse (1986) report that healthy individuals are unlikely to have a consensual prototype for complex illnesses such as cancer. They suggest that *'...the prototypes are likely to be idiosyncratic capitalizing on chance aspects of the persons experience'* (pg 109). Work on breast cancer patients also suggests that disparate illness perceptions are held depending on experience with the disease (Leventhal et al. 1986, Buick et al. 1997). Representations of breast cancer held by women at increased risk of the disease are also likely to vary depending on their experiences in their family. It was therefore hypothesised that experiences of breast cancer in the family will be associated with perceptions of the disease in women at increased risk (H5).

Differences in patterns of illness perceptions (illness representation clusters) are firstly investigated followed by tests of associations between experience items and dimensions of illness perceptions as well as causal items. A number of predictions were made concerning association between experiences in the family and illness perceptions in the increased risk sample. These are outlined as follows.

- Women who reported their experience as more traumatic would perceive breast cancer as more negative. (Specifically have higher scores on the identity, consequences and emotional representations subscales).
- Women who had suffered bereavement from breast cancer or reported that the disease had made greater changes to their life would perceive breast cancer as more negative. (Specifically have higher scores on the timeline

acute/chronic, consequences and emotional representations subscales and lower scores on the personal and treatment control subscales).

- Women who reported that their relative coped well with the disease or reported more positive aspects of the experience would perceive breast cancer as more positive. (Specifically have higher scores on the personal control subscale and lower scores on the consequences subscale).
- Women who had experienced a recent diagnosis of breast cancer in their family were predicted to hold greater emotional representations of the disease and believe more strongly in causal items.

8.1.3 Illness perceptions and experience of breast cancer in the general population: Exploratory analysis.

The third aim of this chapter was to explore the impact of experience of breast cancer on illness perceptions in women in the general population. In line with the mediation model it was predicted that women in the general population with experience of the disease in family or friends would hold more negative perceptions of the illness in terms of higher scores on the identity, timeline acute, consequences and emotional representations subscales (H6).

8.2 MEASURES AND SAMPLES

Data for this analysis was derived from the cross-sectional questionnaire study. The design and procedure were previously described in Chapter 4. The samples reported in this study included: Increased risk sample, general population sample and control sample. These samples were described in Chapter 5.

The measures utilised in this analysis were:

- Experience questionnaire (increased risk sample only) (see Chapter 6, Part 2, sections 6.3-6.8)
- Experience items for the general population sample (see Chapter 4, section 4.4.2b, page 105)
- IPQ-R adapted to assess healthy women's perceptions of breast cancer (see Chapter 6, Part 3, section 6.9-6.15)

8.3 STATISTICAL METHODS

8.3.1 Comparing samples

Differences between samples on the illness representation clusters are assessed with chi-square test and differences on the IPQ-R subscales and causal items are tested with independent t-tests.

8.3.2 Assessing relations between experience and illness perceptions within each sample.

Chi-square tests are used to assess differences in categorical experience measures between the clusters and t-tests are used to determine differences in continuous experience variables between the clusters.

Associations between IPQ-R subscales, causal items and experience measures are assessed with either t-tests or Pearsons correlations depending on the nature of the experience variable (categorical or continuous). Where multiple test are conducted to explore associations with causal items the significance level is adjusted to $p<0.01$ accordingly.

8.4 RESULTS

8.4.1 Comparing illness perceptions between samples with different experiences of breast cancer.

8.4.1a Illness representation clusters

A greater proportion of women in the increased risk sample (Sample A) were classified in the 'negative representation' cluster (n= 51, 52%) than women in the control sample (Sample C) (n= 25, 30.5%); (chi-square= 8.501, df= 1, p= 0.004).

8.4.1b IPQ-R subscales

The mean scores on the IPQ-R subscales for the increased risk and general population samples were provided in Chapter 6 (see Tables 6.12 and 6.13, pages 175&176). Table 8.1 reports the differences in illness perceptions between the increased risk sample (Sample A) and control sample of women in the general

population without any experience of the disease (Sample C). Women at increased risk had a more coherent understanding of breast cancer than the control sample ($p<0.05$). Women at increased risk of breast cancer also had stronger emotional representations of breast cancer and believed the disease to hold greater consequences than controls ($p<0.05$).

Table 8.1- Differences in illness perceptions between the increased risk (A) and control sample (C).

IPQ-R subscale		**Increased risk sample (A)**	**Control sample (C)**	**df**	**t**	**P**
Illness coherence	n	111	94	203	6.65	<0.001
	Mean	2.5	3.1			
	sd	0.69	0.64			
Emotional representations	n	105	91	194	2.16	0.032
	Mean	3.3	3.1			
	sd	0.69	0.67			
Consequences	n	109	94	201	1.97	0.05
	Mean	3.9	3.8			
	sd	0.38	0.41			

8.4.1c Causal beliefs

On average women at increased risk of breast cancer agreed with 6.2 (sd= 3.20) of the causal items (rated agree/strongly agree) compared to 5.5 (sd= 3.41) in the control sample. This difference was not significant (t= 1.48, df= 215, p= 0.140).

Differences in rating scores for individual causal items are provided in Table 8.2. Women at increased risk were more likely to agree that heredity, ageing and hormonal factors were causes of breast cancer than controls ($p<0.05$). They were also more likely to disagree that a germ or virus or poor medical care in the past were causes of breast cancer than controls ($p<0.05$).

Table 8.2- Differences in causal items between the increased risk (A) and control sample (C).

Causal item		**Increased risk sample (A)**	**Control sample (C)**	**df**	**t**	**p**
Heredity- it runs in the family	**n**	117	99	214	4.15	<0.001
	Mean	4.6	4.2			
	sd	0.49	0.64			
Poor medical care in the past	n	117	96	211	2.85	0.005
	Mean	2.3	2.6			
	sd	0.92	0.84			
Ageing	n	116	98	212	2.19	0.03
	Mean	3.3	3.0			
	sd	0.92	0.91			
Hormonal	n	117	98	213	2.19	0.03
	Mean	3.7	3.5			
	sd	0.75	0.81			
A germ or virus	n	116	96	210	1.99	0.048
	Mean	2.1	2.4			
	sd	0.93	0.83			

8.4.2 Experience and illness perceptions in the increased risk sample

8.4.2a Differences in experience between Illness representation clusters

Table 8.3 shows differences in experience items between women in the positive and negative representation clusters. Participants in the 'negative representation' cluster were more likely to have had a more recent breast cancer related bereavement of an index relative or a relative they had known personally than those in the 'postive representation' cluster ($p<0.05$). Participants in the negative cluster were more likely to report that their life plans had changed because of the risk of breast cancer in the family and were less likely to report that their experiences had been positive ($p<0.05$). Women in the 'negative representation' cluster were less likely to report that they resembled their index relative ($p<0.05$).

Table 8.3- Differences in experience between women in the 'positive representation' and 'negative representation' clusters.

Experience item		**Negative cluster**	**Positive cluster**	**df**	**t**	**p**
Recency of bereavement of index relative from breast cancer (if any) (months)	n Mean sd	32 161.6 124.61	26 245.5 143.33	56	2.39	0.02
Experience item 10. *'How much do you feel that your life plans have changed because of the risk of cancer in your family?'*	n Mean sd	49 2.1 1.18	47 1.6 0.90	94	2.64	0.01
Recency of bereavement of a personally known relative from breast cancer (if any) (yrs)	n Mean sd	38 14.1 11.10	34 20.4 12.9	70	2.21	0.031
'Resemblance' subscale	n Mean sd	48 5.2 1.91	47 6.1 2.17	93	2.14	0.035
Experience item 12 *'To what extent have your experiences been positive?'*	n Mean sd	49 2.2 0.97	47 2.7 1.25	94	2.11	0.038
'Traumatic experience' subscale	n Mean sd	46 12.3 1.84	46 11.4 2.61	90	1.75	0.083
Experience item 11 *'Do you feel that your experiences have brought the family closer together?'*	n Mean sd	49 2.4 1.20	47 2.8 1.30	94	1.73	0.087

8.4.2b Significant associations between IPQ-R subscales and experience variables

The following sections describe significant associations between each of the dimensions of illness perceptions assessed by the IPQ-R and experience variables. Table 8.4 provides a summary of the significant correlations ($p<0.05$).

Identity

Respondents who associated more symptoms with breast cancer were more likely to have lost their index relative from breast cancer more recently (r=-.27, p= 0.023, n=

71) and personally knew more relatives who had suffered from breast cancer (r=.18, p= 0.05, n= 116). Individuals with a higher identity score were more likely to report that their experiences brought the family closer together (r= .19, p= 0.041, n= 115).

Timeline acute/chronic

Individuals who perceived breast cancer as more chronic and long-lasting were likely to have lost more relatives from breast cancer (r= .25, p= 0.009, n= 110). Women who perceived breast cancer as long in duration were likely to have lost their index relative from breast cancer more recently (r= -.32, p= 0.009, n= 66) and to have experienced a breast cancer related bereavement of a personally known relative more recently (r= -.29, p= 0.01, n= 80). They were also likely to have reported greater changes in their life plans because of the risk of breast cancer (r= .19, p= 0.045, n= 109).

Consequences

Individuals who perceived breast cancer as holding more serious consequences were less likely to report their experiences had been positive (r= -.36, p<.001, n= 107) and more likely to report that their relative's illness was traumatic (r= .28, p= 0.004, n= 102). They were also likely to have lost their index relative from breast cancer more recently (r= -.26, p= 0.037, n= 66).

Personal control

Individuals who reported less belief in personal control over breast cancer reported greater changes in life plans because of the risk of cancer in the family (r= -.24, p= 0.014, n= 109) and were more likely to report that the illness had brought the family closer together (r= .28, p= 0.004, n= 109).

Treatment control

Participants who had lost one or more personally known relative because of breast cancer reported significantly less belief in treatment control of breast cancer (n= 83, mean= 3.6, sd = 0.53) than participants who had not lost a relative to breast cancer (n= 28, mean= 3.8, sd= 0.45); (t= -2.42, df =109, p= 0.017). Individuals who reported less belief in treatment control of breast cancer were likely to have suffered

bereavement more recently (r= .29, p= 0.009, n= 81) and to have lost their index relative more recently (r= .34, p= 0.00, n= 67). Women with less belief in the treatment control of breast cancer were also less likely to report their experiences had been positive (r= .30, p= 0.002, n= 110) and more likely to feel that their life plans had changed because of the risk of cancer in their family (r=-.26, p= 0.007, n= 110).

Illness coherence

Individuals with a coherent understanding of breast cancer were more likely to report that they resembled their index relative (r= -.22, p= 0.022, n= 108).

Emotional representations

Individuals with stronger emotional representations of breast cancer were more likely to report that their index relative's illness was traumatic (r= .39, p<.001, n= 98) and that their role in the family (r= .20, p= 0.04, n= 103) and life plans had changed because of breast cancer (r= .22, p= 0.027, n= 103).

Table 8.4 - Summary of significant correlations between IPQ-R subscales and experience variables in the increased risk sample.

Subscale	Experience item	Pearsons Correlation Coefficient	n	p
Identity	Recency of bereavement of index relative	-.27	71	0.023
	Do you feel that your experiences have brought the family closer together?	.19	115	0.05
	Number of relatives personally known to have suffered from breast cancer	.18	116	0.041
Timeline acute/ Chronic	Recency of bereavement of index relative	-.32	66	0.009
	Recency of bereavement of a personally known relative from breast cancer	-.29	80	0.01
	Number of personally known relatives who have died from breast cancer	.25	110	0.009
	How much do you feel your life plans have changed because of the risk of cancer in your family?	.19	109	0.04
Consequences	*To what extent have your experiences been positive?*	-.36	107	0.01
	Trauma	.28	102	0.004
	Recency of bereavement of index relative	-.26	66	0.037
Personal control	*Do you feel that your experiences have brought the family closer together?*	.28	109	0.004
	How much do you feel your life plans have changed because of the risk of cancer in your family?	-.24	109	0.014
Treatment control	Recency of bereavement of index relative	.34	67	0.001
	To what extent have your experiences been positive?	.30	110	0.002
	Recency of bereavement of a personally known relative from breast cancer	.29	81	0.009
	How much do you feel your life plans have changed because of the risk of cancer in your family?	-.26	110	0.007
Illness coherence	Resemblance	-.22	108	0.022
Emotional representations	Trauma	.39	98	0.001
	How much do you feel your life plans have changed because of the risk of cancer in your family?	.22	103	0.027
	Do you feel that your role in the family has changed because of your experiences of breast cancer?	.20	103	0.04

8.4.2c Associations between causal beliefs and experience in the increased risk sample.

8.4.2.c(i) Number of causal beliefs and experience

Participants who had a relative they personally knew diagnosed within 5 years agreed with fewer causal items (n= 38, mean= 5.3, sd= 2.66) than those who had not had a relative diagnosed within 5 years (n= 74, mean= 6.6, sd= 3.36); (t= 2.06, df= 110, p= 0.042).

The number of items participants agreed were causes of breast cancer was significantly positively correlated with the following experience items:

- 'Traumatic experience' subscale (r= .25, p= 0.01, n= 110)
- 'Resemblance' subscale (r= .21, p= 0.029, n= 114)

The number of items participants agreed were causes of breast cancer was significantly negatively correlated with:

- Experience item 12: '*To what extent have your experience been positive?*' (r= -.28, p= 0.003, n= 115)
- Relatives age at diagnosis (r=-.22, p= 0.017, n= 115)
- Participants age at relatives diagnosis (r= -.22, p= .016, n= 117)

8.4.2.c(ii) Individual causal items and experience variables

A number of differences in causal items were found between categorical experience variables. Where these differences were significant ($p<0.05$) mean values for the causal items are provided in Tables (Table 8.5-8.8).

Women who had lost at least one relative from breast cancer were more likely to agree that patients mental attitude and emotional state were causes of breast cancer compared to individuals who had not lost a relative from breast cancer ($p<0.05$). These differences are demonstrated in Table 8.5.

Table 8.5- Breast cancer related bereavement and causal beliefs in the increased risk sample.

Causal item		**No relative died from breast cancer**	**At least one relative has died from BC**	**df**	**t**	**p**
Patients mental attitude	n	28	88	114	2.92	0.004
	Mean	2.1	2.7			
	sd	0.57	0.98			
Emotional state	n	28	87	113	2.65	0.009
	Mean	2.1	2.6			
	sd	0.65	0.88			

Women who reported that a relative they personally knew had been diagnosed with breast cancer in the past 5 years were less likely to agree with a number of causal items (Family problems, diet or eating habits, patients mental attitude, overwork, alcohol and smoking) than women who did not have a relative diagnosed with breast cancer in the past 5 years ($p<0.05$) (Please see Table 8.6).

Table 8.6- Recent diagnosis of breast cancer in the family and causal beliefs in the increased risk sample.

Causal item	Relative diagnosed within 5 yrs		No diagnosis within 5 years	Df	t	p
Family problems	n	38	72	108	2.48	0.015
	Mean	2.2	2.7			
	sd	0.82	0.89			
Diet or eating habits	n	38	74	110	2.36	0.02
	Mean	3.0	3.5			
	sd	1.12	0.89			
Patients mental attitude	n	38	74	110	2.24	0.027
	Mean	2.2	2.7			
	sd	0.91	0.93			
Overwork	n	38	74	110	2.11	0.037
	Mean	2.2	2.5			
	sd	0.75	0.74			
Alcohol	n	38	73	109	2.06	0.041
	Mean	2.4	2.8			
	sd	0.85	0.88			
Smoking	n	36	74	108	2.01	0.047
	Mean	3.1	3.5			
	sd	1.17	0.89			

Women who chose their mother as their index relative were more likely to agree that 'Alcohol' and 'Smoking' were causes of breast cancer ($p<0.05$) (Please see Table 8.7).

Table 8.7- Relation to the Index relative and causal beliefs in the increased risk sample.

Cause item		Index relative was mother	Index relative was not mother	df	t	p
Alcohol	n	80	36	114	2.60	0.01
	Mean	2.8	2.4			
	sd	0.92	0.79			
Smoking	n	81	34	113	2.49	0.014
	Mean	3.6	3.1			
	sd	0.79	1.01			

Participants who reported that their index relative had died from breast cancer were more likely to agree that patient's mental attitude was a cause of breast cancer and less likely to agree that heredity was a cause of the disease than women whose index relative was still alive ($p<0.05$) (See Table 8.8).

Table 8.8- Index relative status and causal beliefs in the increased risk sample.

Cause item		**Index relative had died from breast cancer**	**Index relative is alive**	**df**	**t**	**p**
Patients Mental attitude	n	71	39	108	2.64	0.01
	Mean	2.7	2.2			
	sd	0.93	0.84			
Heredity- it runs in the family	n	71	39	108	2.61	0.01
	Mean	4.5	4.7			
	sd	0.50	0.46			

There were a number of correlations between experience items and causal beliefs although the strength of the correlations were not particularly high. Those reaching significance of $p<0.01$ are reported here.

The number of relatives personally known by the participant who had suffered from breast cancer was positively correlated with belief that '*Heredity-it runs in the family*' is a cause of breast cancer ($r= .24$, $p= 0.009$, $n= 117$).

Participants with higher scores on the experience subscale 'Trauma' were more likely to agree with the causal item *'Hormonal'* ($r= .27$, $p= 0.004$, $n= 110$). Participant with higher scores on experience item 9 *'Do you feel that your role in the family had been changed because of your experiences of breast cancer?'* were more likely to agree with the causal item *'Diet or eating habits'*($r= .27$, $p= 0.004$, $n= 115$).

Participant who score higher on subjective experience item 12 '*To what extent have your experiences been positive'* were less likely to agree with the causal *'Altered immunity'* ($r= -.30$, $p= 0.001$, $n= 115$).

8.4.3 Experience and illness perceptions in the general population sample (Sample B&C)

8.4.3a Differences in experience between Illness representation clusters

There were no significant differences between the illness representation clusters and experiences of breast cancer in the general population sample. There was a trend for women in the 'negative representation' cluster to be more likely to have had a recent experience of breast cancer in family or friends (26.9%) compared to participants in the 'positive representation' cluster (16.7%); (chi square= 3.23, df= 1, p= 0.072).

8.4.3b Differences in IPQ-R subscales between women with different experiences of breast cancer in the general population.

Women who reported different experiences of breast cancer in the general population sample showed differences on illness perceptions as assessed by the IPQ-R. These differences are described for each dimension as follows.

Consequences

Women with any experience of breast cancer, a recent experience of breast cancer or 'other' experiences of breast cancer believed the disease to hold more consequences than the control sample of women with no experience of the disease at all (Sample C) ($p<0.05$) (See Table 8.9). Women with a recent experience of breast cancer in friends of family believed the disease to hold more consequences (n= 49, mean= 4.1, sd= 0.40) than women with no recent experience (n= 190, mean= 3.9, sd= 0.43); (t=3.36, df= 237, p= 0.001).

Table 8.9- Differences in perceptions of the consequences of breast cancer for women in the general population with different experience of the disease.

Experience of breast cancer		**Experience of breast cancer**	**Control sample (C)**	**df**	**t**	**p**
A member of the family or close friend has suffered from breast cancer recently	n Mean sd	49 4.1 0.40	94 3.8 0.41	141	3.49	0.001
'Other' experiences of breast cancer.	n Mean sd	85 4.0 0.46	94 3.8 0.41	177	2.34	0.021
Any experience of breast cancer	n Mean sd	146 3.9 0.45	94 3.8 0.41	238	2.01	0.045

Timeline acute/chronic

Women who had had a recent experience of breast cancer believed the disease to be more chronic and long-lasting (n= 50, mean= 3.4, sd= 0.59) than women without a recent experience (n= 192, mean= 3.2, sd= 0.50); (t= 2.04, df= 240, p= 0.042).

Illness coherence

Participants who reported any experience of breast cancer (including that a relative that had suffered from breast cancer, a recent experience of breast cancer, experience of breast cancer at work or 'other' experiences of breast cancer) had a more coherent understanding of breast cancer than controls ($p<0.05$) (See Table 8.10). In addition, women who had experience of breast cancer at work had a more coherent understanding of breast cancer than women who did not have experience of breast cancer at work (n= 214, mean= 3.0 sd= 0.69); (t= 2.61, df= 241, p= 0.01).

Table 8.10- Differences in illness coherence for women in the general population with different experience of breast cancer.

Experience of breast cancer		**Experience of breast cancer**	**Control sample (C)**	**df**	**t**	**p**
Experience of breast cancer at work	n Mean sd	29 2.6 0.75	94 3.1 0.64	121	3.36	0.001
Any experience of breast cancer	n Mean sd	151 2.8 0.73	94 3.1 0.64	243	2.75	0.006
'Other' experiences of breast cancer	n Mean sd	88 2.8 0.72	94 3.1 0.64	180	2.47	0.014
A relative had suffered from breast cancer	n Mean sd	60 2.82 .73	94 3.1 0.64	152	2.25	0.026
A member of the family or close friend has suffered from breast cancer recently	n Mean sd	50 2.8 0.81	94 3.1 0.64	142	2.13	0.035

Emotional representations

Women with experience of breast cancer at work reported significantly lower emotional representations of breast cancer than women with no experience of breast cancer at work (n= 209 mean= 3.2, sd= 0.66); (t= 3.84, df= 236, p<.001) and women with no experience of the disease (p<0.05) (See Table 8.11). Women with a recent experience of breast cancer had stronger emotional representations of the disease (n= 50, mean = 3.3, sd= 0.72) than women without a recent experience of breast cancer (n= 189, mean = 3.1 sd= 0.66); (t= 2.56, df= 237, p= 0.011).

Table 8.11- Differences in emotional representations for women in the general population with different experience of breast cancer.

Experience of breast cancer		Experience of breast cancer	Control sample (C)	df	t	p
Experience of breast cancer at work	n Mean sd	29 2.67 .65	91 3.12 .67	118	3.17	0.002

8.4.3c Associations between causal beliefs and experience in the general population sample

8.4.3c(i) Any experience of breast cancer

Individuals with any experience of breast cancer were significantly more likely to agree that heredity, chance, ageing and hormonal factors were causes of breast cancer than the control sample (C) with no experience of breast cancer ($p<0.05$) (See Table 8.12).

Table 8.12- Experience of breast cancer in the general population sample and causal beliefs.

Cause item		Any experience of breast cancer	Control sample (C)	df	t	p
Hormonal	n Mean sd	154 3.8 0.73	98 3.5 0.81	250	3.04	0.003
Heredity-it runs in the family	n Mean sd	156 4.5 0.62	99 4.2 0.64	253	2.62	0.009
Chance or bad luck	n Mean sd	152 3.3 1.13	97 3.0 1.04	247	2.18	0.03
Ageing	n Mean sd	154 3.3 0.93	98 3.0 0.91	250	2.18	0.031

8.4.3.c(ii) Experience of breast cancer in the family

Participants who reported that a relative had suffered from breast cancer were significantly more likely to agree that hormonal factors were a cause of breast cancer than controls with no experience of the disease at all ($p<0.05$) (See Table 8.13).

Table 8.13- Recent experience of breast cancer in the general population sample and causal beliefs.

Cause item		**A relative had suffered breast cancer**	**Control sample (C)**	**df**	**t**	**p**
Hormonal	n	64	98	160	2.25	0.026
	Mean	3.7	3.5			
	sd	0.67	0.81			

8.4.3c(iii) Recent experience of breast cancer in friends or family

Participants who reported a recent experience of breast cancer agreed with more causes (mean= 7.4, sd= 3.3, n= 51) than those without recent experience (mean= 5.7, sd= 3.37, n= 204); (t= 3.31, df= 253, p= 0.001) and those with no experience (Sample C)(t= 3.20, df= 149, p= 0.002).

Participants who had had a recent experience of breast cancer in friends or family were significantly more likely to agree that heredity, hormonal factors, patients emotional state, ageing, family problems, patients mental attitude and stress were causes of breast cancer than women without any experience of the disease (Sample C) ($p<0.05$) (See Table 8.14).

Table 8.14- Recent experience of breast cancer in the general population sample and causal beliefs.

Cause item		A family member of close friend has suffered from breast cancer recently	Control sample (C)	df	t	p
Heredity – it runs in the family	n Mean sd	51 4.7 0.48	99 4.24 0.64	148	3.97	0.000
Hormonal	n Mean sd	50 3.9 0.70	98 3.6 0.81	146	2.97	0.004
Emotional state eg feeling down, lonely, anxious, empty	n Mean sd	49 2.9 0.85	97 2.5 0.84	144	2.92	0.004
Ageing	n Mean sd	50 3.4 0.86	98 3.0 0.91	146	2.58	0.011
Family problems	n Mean sd	49 2.9 0.89	97 2.54 .83	144	2.57	0.011
Patients mental attitude eg thinking about life negatively	n Mean sd	49 2.9 0.89	96 2.5 0.83	143	2.45	0.016
Stress or worry	n Mean sd	50 3.5 0.79	97 3.2 0.91	145	2.27	0.025

8.4.3c(iv) Experience of breast cancer at work

Those with experience of breast cancer at work were more likely to rate ageing, patients personality and hormonal factors as causes of breast cancer than women with no experience of breast cancer at all ($p<0.05$) (See Table 8.15).

Table 8.15- Experience of breast cancer at work and causal beliefs in the general population sample.

Cause item		Experience of breast cancer at work	Control sample (C)	df	t	p
Ageing	n	30	98	126	2.59	0.011
	Mean	3.5	3.0			
	sd	0.97	0.91			
Patient's personality	n	28	97	123	2.54	0.012
	Mean	2.6	2.2			
	sd	0.99	0.66			
Hormonal	n	30	98	126	2.55	0.012
	Mean	3.9	3.5			
	sd	0.57	0.81			

8.4.3c(v) 'Other' experiences of breast cancer

Those who reported 'other' experiences of breast cancer were significantly more likely to agree that heredity and hormonal factors were causes of breast cancer and less likely to agree that pollution was a cause of breast cancer compared to those with no experience at all ($p<0.05$) (See Table 8.16).

Table 8.16 'Other' experiences of breast cancer and causal beliefs in the general population.

Cause item		'Other' experience of breast cancer	Control sample (C)	df	t	p
Heredity- it runs in the family	n	90	99	187	2.59	0.01
	Mean	4.5	4.2			
	sd	0.60	0.64			
Pollution in the environment	n	89	97	184	2.25	0.026
	Mean	2.8	3.1			
	sd	0.89	0.83			
Hormonal	n	89	98	185	1.99	0.048
	Mean	3.7	3.5			
	sd	0.82	0.81			

8.5 SUMMARY AND DISCUSSION

Overall the results reported in this chapter have met a number of theoretically driven predictions. Women at increased risk of breast cancer hold different perceptions of the disease than women without experience of breast cancer in their social environment. Women at increased risk have stronger emotional representations of breast cancer and also hold different cognitive representations of the disease than women with no experience of the disease. Perceptions of breast cancer in the increased risk sample are associated with specific aspects of experiences of breast cancer in the family. In addition, women in the general population with different experiences of breast cancer hold different representations of the disease.

8.5.1 Differences in illness perceptions between samples with different experiences of breast cancer.

As predicted, women in the increased risk sample were more likely to hold an overall negative pattern of illness representations than women without experience of the disease. Women at increased risk of breast cancer perceived breast cancer as holding greater consequences and held stronger emotional representations of the disease than controls. Women in the increased risk sample also reported that they had a more coherent understanding of the disease than women without any experience of breast cancer.

These results suggest that women at increased risk were not minimizing their threat as shown in other screening situations (Jemmott et al. 1986, Croyle et al. 1997, Rimes et al. 1999). Threat minimization has been proposed as most likely when the risk is uncertain and there are limited options for control over risk (Ditto et al. 1988). A genetic predisposition to breast cancer would therefore appear to be a likely clinical situation for threat minimization to occur. The lack of this effect raises the possibility that genetic based information impedes these defensive processes or that individuals with experience of the disease in question are less likely or unable to engage in threat minimisation. However, this study was not designed to directly assess threat minimization process and further tests of this mechanism are required.

The differences between the samples were not to the extent that was predicted. No difference in perception of the duration of breast cancer (timeline acute/chronic subscale) was found between the samples. This may be due to variable duration of illness that women at increased risk of breast cancer were exposed to. It may also be due to poor psychometric properties of this subscale demonstrated by lower reliability (see section 6.19.1, pages 171-174). It was surprising that the samples did also not show any differences in perceptions of control of breast cancer. It was also surprising that women at increased risk of breast cancer believed in a similar number of causal factors as did controls. This may be due to the large number of factors potentially implicated in breast cancer risk and widespread information about risk factors available to women in the general population (see Chapter 1). However women in the increased risk sample did believe more strongly in a number of medically correct causes as predicted (eg heredity, ageing, hormonal factors) compared to the control sample.

There was concern that women at increased risk of breast cancer may be fatalistic about the disease (French et al. 2001, Turnquist et al. 1988). However, women in the increased risk sample showed stronger beliefs in some controllable items (eg diet) and were less likely to believe in uncontrollable items (eg poor medical care in the past) than controls. This suggests that those women facing an increased risk of breast cancer because of their family history of the disease were still able to believe in controllable causes and were not deterministic about their risk.

8.5.2 Illness perceptions and experience of breast cancer in the increased risk sample.

In the increased risk sample experiences of breast cancer in the family were associated with the overall pattern of beliefs represented by the clusters. As predicted participants in the 'negative representation' cluster were more likely to report negative experiences of breast cancer (ie a more recent bereavement of a relative from breast cancer, greater changes in life plans because of the risk of breast cancer in the family and less positive aspects of the experience). It was unexpected that women in the 'negative representation' cluster were *less* likely to report that they resembled their index relative. This may possibly reflect attempts at threat

minimization within this sample. Women who feel they resemble their index relative judge the risk as less threatening than women who do not feel they resemble their index relative. Further work would be required to explore this issue in regards to beliefs about inheritance and genetics.

All dimensions of illness perceptions were associated with aspects of experience and the vast majority of associations were in the expected direction (see section 8.1.2, pages 230-231). This suggests that living with a family history of breast cancer has a wide impact on representations of the disease. As predicted women who reported their experiences as more traumatic believed breast cancer to hold more consequences and held stronger emotional representations of the disease. Women who had lost their index relative more recently associated more symptoms with breast cancer, believed the disease to be long lasting with severe consequences and were less likely to believe in the treatment control of breast cancer. Recent or traumatic experiences are likely to create accessible images of breast cancer in mind (see 3.2, page 73) and therefore have a strong impact on the development of representations of the disease.

Women who reported that breast cancer had changed their life plans believed the illness to be long-lasting, held strong emotional representations of the disease and were less likely to believe in the personal or treatment control of breast cancer. This may indicate that women who have felt the consequences of breast cancer on their own lives develop stronger representations of the disease. Positive experiences were shown to have a beneficial effect on illness representations. Women who reported that their experiences had been more positive were more likely to believe in the treatment control of breast cancer and perceived the disease as holding less consequences and women who reported that the experience had brought the family closer together were more likely to believe in personal control of the disease.

A few associations occurred that had not been predicted. Women who reported that their experiences had brought the family closer together associated more symptoms with breast cancer. It is possible that this may reflect greater contact with the index relative or increased awareness and discussion about symptoms of the disease.

Women who reported that they resembled their index relative had a more coherent understanding of breast cancer. Although the majority of predicted associations between experiences of breast cancer in the family and illness perceptions were met no associations were found between how well the participant perceived their relative to cope with breast cancer and illness perceptions. This may suggest that a relative's personal response and ability to cope with breast cancer may not influence a healthy relatives perception of the disease or that a single item is inadequate to grasp this construct.

Experience of breast cancer in the family was widely associated with causal beliefs about the disease. In contrast to predictions women who had had a diagnosis in their family within 5 years believed in fewer of the causal items provided on the causal checklist of the IPQ-R and were *less* likely to agree with a number of the individual causal items. However, individuals who had lost a relative from breast cancer were more likely to agree with a number of causal items. This may reflect women searching for causal explanations for the disease following bereavement rather than diagnosis. Respondents in the increased risk sample who had lost a relative to breast cancer were more likely to agree with causal factors implying patient characteristics were important causes of breast cancer (e.g. patients mental attitude). In addition women who had lost their index relative to breast cancer were less likely to believe that heredity was a cause of breast cancer. Turnquist et al. (1988) suggested that healthy relatives of patients often attribute the illness to the patient's personal characteristics as a means to reduce responsibility for having caused or concerns about possible prevention of the disease. In this sample of women at increased risk of breast cancer, attributing a relatives illness to personal characteristics rather than heredity may also reflect a defensive strategy to reduce feelings of vulnerability to the disease.

8.5.3 Illness perceptions and experience of breast cancer in the general population sample.

Exploratory analysis revealed that there was no significant difference in the overall pattern of beliefs held by women with different experiences of breast cancer in this sample. However a number of individual subscales and causal items were associated

with experience in this sample. Consistent with predictions and the results from the increased risk sample, women in the general population with more experience of breast cancer believed the disease to hold stronger consequences. This effect was particularly strong for women with a recent experience of breast cancer in friends or family. Recent experience of breast cancer was also associated with the belief that breast cancer was a long-lasting illness and stronger emotional representations of breast cancer. The illness coherence subscale was also associated with experience of breast cancer in the general population sample. Women who reported any experience of breast cancer showed a more coherent understanding of the disease than women without any experience of breast cancer. This suggests that experiences of breast cancer in women in the general population may have beneficial as well as negative effect on representations of the disease.

Experience of breast cancer at work appeared to have a buffering effect and was associated with a lower emotional representation of breast cancer and a more coherent understanding of the disease. This indicates that exposure to breast cancer per se does not have uniform effects on illness perceptions but that the qualitative nature of the exposure determines its impact on perceptions of and emotional response to the disease in individuals not at risk.

CHAPTER 9

ILLNESS PERCEPTIONS AND DISTRESS

ILLNESS PERCEPTIONS AND DISTRESS

9.1 AIMS AND RATIONALE

Analysis reported in Chapter 7 and 8 examined two elements of the mediation model (Figure 4.1, page 100) - associations between experience of breast cancer with levels of distress and illness perceptions. A number of associations were identified in both sets of analyses. The main aim of the following analysis was to address the final section of the mediation model (Figure 4.1, objective 4) and assess the contribution of illness perceptions to levels of general and cancer specific distress in women at increased risk of breast cancer.

Large variations in levels of distress across individuals have been reported in women at increased risk of breast cancer (Kash et al. 1992, Gagnon et al. 1996, Hopwood et al. 1998, Cull et al. 1999, Watson et al. 1999) (see Chapter 1, section 1.4, page 48). However, theoretically driven research aimed at understanding factors accounting for variability in distress has been limited. Research based on the SRM has indicated that illness perceptions are associated with patient's emotional response to a range of chronic illnesses including CFS, rheumatoid arthritis, osteoarthritis, multiple sclerosis, psoriasis and breast cancer (Heijmans 1988, Murphy et al. 1999, Orbell et al. 1998, Schiaffino et al. 1998, Fortune et al. 2000, Buick 1997) (See Chapter 4, section 3.4.5c, page 88). Women's emotional response to breast cancer *risk* may also be associated with representations of the disease.

9.1.1 Associations between illness perceptions and levels of general and cancer specific distress in each sample: Predictions.

It was hypothesised that perceptions of breast cancer would be significantly associated with levels of general and cancer specific distress in women with a significant family history of breast cancer (H7). The first aim of this analysis was to examine associations between a) patterns of beliefs as represented by the illness representation clusters; b) dimensions of perceptions as assessed by the IPQ-R subscales and c) causal items, with levels of general and cancer specific distress. It was predicted that women in the increased risk sample with more negative representations of breast cancer would show higher levels of distress than women with more positive perceptions of the disease. Higher levels of distress were expected

in women in the 'negative representation' cluster compared to the 'positive representation' cluster. Scores on the IPQ-R subscales (in parentheses) were expected to be correlated with levels of distress. Specifically higher levels of distress were predicted for women who:

- Associated more symptoms with breast cancer (Identity)
- Perceived the disease as long lasting (Timeline acute/chronic)
- Perceived the disease as holding greater consequences (Consequences)
- Did not believe in the personal or treatment control of breast cancer (Personal control, Treatment control)
- Did not have a coherent understanding of the disease (Illness coherence)
- Had strong emotional representations of breast cancer (Emotional representations)

Stronger associations were expected between illness representations and cancer specific distress compared with measures of general distress. Exploratory analysis was also conducted to examine associations between causal items and distress in these samples.

The associations between illness perceptions and levels of distress in the control sample was examined for comparison. It was predicted that fewer associations between illness representations and distress measures would be found in this sample and the associations would be weaker than those in the increased risk sample.

9.1.2 Predicting levels of general and cancer specific distress in the increased risk sample: Exploratory analysis

The second aim of the analysis reported in this Chapter was to examine the predictive value of illness perceptions in understanding variations in levels of distress across individuals. Initially multiple regression analysis was conducted to determine how much variability in distress was explained by illness representations alone. It was predicted that emotional representations would account for a large proportion of variance in cancer specific worry due to conceptual overlap between these scales. Analysis was therefore conducted twice, once with emotional representations

included in the model, and again with emotional representations omitted from the model in order to derive a clinically useful result.

The results reported in Chapter 8 indicated that the experience items accounted for a small proportion of the variance in levels of distress. Analysis was therefore conducted including both experiences items and illness perception dimensions in the multiple regression models in order to identify the *best predictors* of distress in women at increased risk of breast cancer.

9.2 MEASURES AND SAMPLES

Data for this analysis were derived from the cross-sectional questionnaire study. The design and procedure were previously provided in Chapter 4. The samples reported in this study included: Increased risk sample (Sample A) and control sample (Sample C). These samples were described in Chapter 5.

The measures utilised in this analysis were:

- General distress (GHQ-30) (see Chapter 4, section 4.4.4a, page 106)
- Cancer specific distress (Cancer worry scale, Impact of Event scale) (see Chapter 4, section 4.4.4b, page 107)
- IPQ-R adapted to assess healthy women's perceptions of breast cancer (see Chapter 6, Part 3, section 6.9-6.15)

9.3 STATISTICAL METHODS

9.3.1 Associations between illness perceptions and distress within each sample

Chi square and T-tests were used to determine any differences in levels of distress between participants in each illness representation cluster. T-tests are also used to determine any differences in illness perceptions and causal items between GHQ 'cases' and 'non cases' and participants who did and who did not complete the Impact of Event Scale in each sample. Correlational analysis was conducted using Pearson Correlation Coefficient to determine associations between illness perceptions, causal items and levels of distress. Given the number of causal items examined the p value for this analysis was adjusted to $p<0.01$.

9.3.2 Predicting levels of distress in the increased risk sample

Multiple regression was used to determine best predictors of general and cancer specific distress in the increased risk sample (Sample A). The background multiple regression and stages of analysis were outlined in Chapter 7 (see section 73.2, page 202Due to conceptual overlap and high associations between the emotional representations subscale of the IPQ-R and cancer specific distress measures, regression models predicting cancer worry were conducted with and without this subscale.

9.4 RESULTS

9.4.1 Associations between illness perceptions and distress in the increased risk sample

9.4.1a General distress

There was a higher proportion of GHQ 'cases' in the 'negative representation' cluster (n=19, 38%) compared with the 'positive representation' cluster (n= 11, 23.4%) but this association was not statistically significant (chi-square= 2.42, df= 1, p= 0.12). Individuals in the two clusters did not differ on GHQ score (t=1.31, df= 95, p= 0.19) ('Negative representation' cluster, n= 50, mean= 28.5, sd= 12.77); ('Positive representation' cluster, n= 47, mean= 25.3, sd= 11.20).

Illness representations were associated with GHQ 'caseness'. GHQ 'cases' scored significantly higher on the identity ($p<0.01$), timeline acute/chronic and emotional representation subscales of the IPQ-R ($p<0.05$) (see Table 9.1). There was also a trend for GHQ 'cases' to score higher on the consequences subscale ($p<0.1$) (see Table 9.1). There were no significant differences between GHQ 'cases' and 'non cases' on the causal items.

Table 9.1- Differences in illness perceptions between GHQ 'cases' and 'non cases' in the increased risk sample.

IPQ-R subscale		GHQ 'case'	'Non case'	df	t	p
Identity	n	36	80	114	3.25	0.002
	Mean	5.4	4.0			
	sd	2.41	2.1			
Timeline acute/ chronic	n	36	74	108	2.54	0.012
	Mean	3.5	3.2			
	sd	0.59	0.47			
Emotional representations	n	31	73	102	2.13	0.035
	Mean	3.6	3.2			
	sd	0.64	0.70			
Consequences	n	34	74	106	1.71	0.089
	Mean	4.0	3.9			
	sd	0.37	0.38			

The correlation matrix (Table 9.3) indicated that a number of dimensions of illness representations were significantly correlated with GHQ score[9]. These were: identity ($p<0.01$); emotional representations ($p<0.01$); timeline acute/chronic and consequences ($p<0.05$). Women with higher levels of distress associated more symptoms with breast cancer, held stronger emotional representations of the disease, believed the disease was longer lasting with greater consequences.

No causal items were significantly correlated with GHQ score ($p<0.01$)

9.4.1b Cancer specific distress

9.4.1b(i) Cancer worry

Participants in the 'negative representation' cluster reported significantly higher levels of cancer worry (mean= 11.5, sd= 2.64) than participants in the 'positive representation' cluster (mean= 9.6, sd= 1.81, n= 47); ($t= 4.15$, $df= 96$, $p<0.001$).

The correlation matrix (Table 9.3) indicates that cancer worry score was strongly correlated emotional representations ($p<0.001$), timeline acute/chronic ($p<0.001$) and

[9] Similar correlation coefficients were found using the Logarithmic transformation of GHQ score

was also significantly correlated with consequences ($p<0.01$) and identity ($p<0.05$). Women with higher levels of cancer worry had stronger emotional representations of the disease, perceived breast cancer to be of longer duration with severe consequences and believed more symptoms to be associated with the disease. There were no significant correlations between the causal items and cancer worry score ($p<0.01$).

9.4.1b(ii) Impact of Event scale

Significantly more women in the 'negative representation' cluster reported they had thought about breast cancer in the previous week and completed the Impact of Event scale (n= 35, 67%) than women in the 'positive representation' cluster (n= 17, 33%) (Chi-square= 10.35, df= 1, p= 0.001). Women in the 'negative representation' cluster scored significantly higher on the intrusion subscale of the measure (n= 34, mean = 8.7, sd= 5.76) than women in the 'positive representation' cluster (n= 17, mean = 4.9, sd= 3.77); (t= 2.42, df= 49, p= 0.019).

Table 9.2 shows differences in illness representations between women who did and did not report to have thought about breast cancer in the previous week. Women who had thought about breast cancer in the previous week and completed the Impact of Event scale showed stronger emotional representations of breast cancer and believed more symptoms to be associated with breast cancer (higher identity score) than women who did not complete the scale ($p<0.05$). There was also a trend for individuals who had completed the scale to have a less coherent understanding of breast cancer and to believe the disease was longer lasting than individuals who did not complete the scale ($p<0.1$). There were no differences on any of the causal items between participants who did and did not complete the Impact of Event scale.

Table 9.2- Differences in illness perceptions between participants in the increased risk sample who had and had not thought about breast cancer in the previous week.

IPQ-R subscale	Had thought about breast cancer in the previous week		Had not thought about breast cancer in the previous week	df	t	p
Emotional representations	n	57	48	103	2.56	0.012
	Mean	3.5	3.2			
	sd	0.63	0.73			
Identity	n	61	56	115	2.12	0.036
	Mean	4.9	4.0			
	sd	2.32	2.10			
Illness coherence	n	59	52	109	1.91	0.058
	Mean	2.6	2.3			
	sd	0.64	0.72			
Timeline acute/chronic	n	60	51	109	1.83	0.071
	Mean	3.4	3.2			
	sd	0.56	0.48			

The correlation matrix (Table 9.3) shows correlations between illness representations with total score; intrusion subscale and avoidance subscale of the Impact of Event scale. The total score and intrusion subscale were strongly correlated with emotional representations ($p<0.001$). The avoidance subscale was also significantly correlated with emotional representations ($p<0.05$) although the relationship was not as strong. The total score and intrusion subscale were significantly negatively correlated with treatment control ($p<0.05$). Intrusion score was also correlated with the timeline acute/chronic subscale ($p=0.05$) Women who reported greater intrusive thoughts about breast cancer had stronger emotional representations of breast cancer, were less likely to believe in the efficacy of treatment to control the disease and perceived the disease as longer in duration.

The intrusion subscale was not significantly correlated any causal item ($p<0.01$)

The avoidance subscale was significantly correlated with the causal items:

- *'Altered immunity'* ($r= .37$, $p= 0.004$, $n= 58$)
- *'A germ or virus'* ($r= .35$, $p= 0.007$, $n= 58$)

Table 9.3- Correlation matrix showing significant correlations (Pearson) between IPQ-R subscales and measures of distress in the increased risk sample ($p<0.1$)

IPQ-R subscale Distress measure	Identity	Timeline acute/ chronic	Consequences	Personal control	Treatment control	Illness coherence	Emotional representations
GHQ	r= .30 p= 0.001 n= 116	r= .21 p= 0.028 n= 110	r= .20 p= 0.035 n= 108				r= .27 p= 0.009 n= 104
Cancer Worry	r= .19 p= 0.045 n= 116	r= .25 p= 0.008 n= 110	r= .20 p= 0.037 n= 109				r= .66 p< 0.001 n= 104
Impact of Event: Total score					r= -.28 p= 0.035 n= 58		r= .48 p< 0.001 n= 55
Impact of Event: Avoidance score							r= .29 p= 0.032 n= 55
Impact of Event: Intrusion score		r= .26 p= 0.05 n= 59		r= .24 p= .066 n= 59	r= -.33 p= 0.007 n= 60		r= .60 p< 0.001 n= 56

9.4.2 Associations between Illness perceptions and distress in the control sample (Sample C)

9.4.2a General distress

There was no difference in the proportion of GHQ 'cases' between women classified in the 'negative representation' or 'positive representation' clusters (chi-square= 1.55, df= 1, p= 0.21). There was also no difference in GHQ score between the two clusters (t= 1.67, df= 80, p= 0.098). Table 9.4 shows the differences in the illness perception dimensions between GHQ 'cases' and 'non cases'. GHQ 'cases' had significantly stronger emotional representations of breast cancer than 'non cases' ($p<0.05$) and showed a trend to perceive breast cancer as holding more severe consequences ($p<0.1$). GHQ 'cases' were likely to agree with more of the causal items (n= 31, mean = 6.7, sd = 3.13) than 'noncases' (n= 67, mean = 5.0, sd= 3.46); (t= 2.28, df= 96, p= 0.023) and were more likely to agree that 'smoking' and 'patients behaviour' were causes of breast cancer ($p<0.05$).

Table 9.4- Differences in illness perceptions between GHQ 'cases' and 'non cases' in the control sample

IPQ-R subscale		GHQ 'case'	Non 'case'	df	t	p
Emotional Representations	n	29	60	87	2.11	0.038
	Mean	3.3	3.01			
	sd	0.55	0.71			
Consequences	n	29	63	90	1.69	0.095
	Mean	3.9	3.8			
	sd	0.41	0.40			
Causal items						
Smoking	n	30	65	93	3.62	<0.001
	Mean	3.9	3.3			
	sd	0.58	0.91			
Patients own behaviour	n	31	65	94	3.00	0.003
	Mean	3.0	2.4			
	sd	0.91	0.81			

The correlation matrix (Table 9.6) indicates that GHQ score was only significantly correlated with emotional representations ($p<0.01$). Women in this sub-sample with

higher levels of general anxiety were likely to have stronger emotional representations of breast cancer.

9.4.2b Cancer specific distress

9.4.2b(i) Cancer worry

There was no difference in cancer worry between women classified in each illness representation cluster (t= 1.53, df= 80, p= 0.13). The correlation matrix (Table 9.6) indicates that cancer worry score was strongly correlated with identity and emotional representations ($p<0.001$). Women with higher levels of cancer worry had stronger emotional representations of the disease and believed more symptoms were associated with breast cancer. There was also a trend for women with higher cancer worry scores to have a less coherent perception of breast cancer ($p<0.1$).

9.4.2b(ii) Impact of Event scale

There was no difference in the proportion of women in each illness representation cluster who reported to have thought about breast cancer in the past week and completed the Impact of Event scale (chi-square= 1.13, df= 1, p= 0.29). For those who completed the scale no differences were found between women in each cluster for the total score (t= .93, df= 19, p= 0.36); avoidance score (t= .86, df= 19, p= 0.40); or intrusion score (t= .91, df= 21, p= 0.38).

Table 9.5 shows differences on IPQ-R subscales between women who reported to have thought about breast cancer in the previous week and completed the Impact of Event scale and those who reported not to have thought about breast cancer in the previous week. Women who reported to have thought about breast cancer in the previous week showed stronger emotional representations of breast cancer ($p<0.05$) and showed a trend to perceive greater personal control over the disease ($p<0.1$).

Table 9.5- Differences in illness perceptions between participants in the control sample who had and had not thought about breast cancer in the previous week.

IPQ-R subscale	**Thought about breast cancer in the previous week**	**Did not think about breast cancer in the previous week.**	**df**	**t**	**p**
Emotional representations	n 23 Mean 3.4 sd 0.52	67 3.03 0.70	88	2.01	0.047
Personal control	n 23 Mean 3.4 sd 0.50	68 3.2 0.39	89	1.79	0.077

The correlation matrix (Table 9.6) indicates that the total score, intrusion and avoidance subscales on the Impact of Event scale were significantly correlated with identity and emotional representations ($p<0.05$). Women with higher levels of intrusive thoughts and avoidance of breast cancer were significantly more likely to have stronger emotional representations of the disease and to associate more symptoms with breast cancer.

Women who believed that 'Patients personality' was a cause of breast cancer were significantly more likely to have a higher total score ($r= .55$, $p= 0.001$, $n= 20$).

Table 9.6- Correlation matrix showing significant correlations (Pearson) between IPQ-R subscales and measures of distress in the control sample ($p<0.1$)

IPQ-R subscale Distress measure	Identity	Timeline acute/chronic	Consequences	Personal control	Treatment control	Illness coherence	Emotional representations
GHQ Likert Score							r= .28 p= 0.009 n= 89
Cancer Worry	r= .39 p< 0.001 n =99					r= -.20 p= 0.055 n= 92	r=.50 p< 0.001 n= 90
Impact of Event: Total	r= .60 p= 0.004 n= 21						r= .55 p= 0.01 n= 21
Impact of Event: Avoidance	r= .57 p= 0.007 n= 21						r= .53 p= 0.014 n= 21
Impact of Event: Intrusion	r= .51 p= 0.014 n= 23						r= .45 p= 0.032 n= 23

9.4.3 Summary of associations between illness perceptions and distress in each sample.

Tables 9.7 and 9.8 provide a summary of the associations between general and cancer specific distress with illness perceptions in each sample. For each measure of distress significant associations with illness representation clusters and illness representation dimensions are reported ($p<0.05$). The strength of associations are also reported (t value or r correlation coefficient) for comparison.

Table 9.7- Summary of associations between general distress and illness representations in each sample

Measure	**Increased risk sample (A)**	**Control sample (C)**
GHQ caseness	Identity (t= 3.25, df= 114, p= 0.002) Timeline acute/chronic (t= 2.54, df= 108, p= 0.012) Emotional representations (t= 2.13, df=102, p= 0.035)	Emotional representations (t= 2.11, df= 87, p= 0.038)
GHQ score	Identity (r= .30, n= 116, p= 0.001) Emotional representations (r= .27, n= 104, p= 0.009) Timeline acute/chronic (r= .21, n= 110, p= 0.028) Consequences (r= .203, n= 108, p= 0.035)	Emotional representations (r= .28, n= 89, p= 0.009)

Table 9.8- Summary of associations between cancer specific distress and illness representations in each sample

Measure	Increased risk sample (A)	Control sample (C)
Cancer worry	Clusters (t= 4.15,df= 96, p<0.001) Emotional representations (r= .66, n= 104, p<0.001) Timeline acute/chronic (r= .25, n= 110, p= 0.008) Consequences (r= .20, n= 109, p= 0.037) Identity (r= .19, n= 116, p= 0.045)	Emotional representations (r = .50, n=90, p<0.001) Identity (r= .39, n= 99, p<0.001)
Completion of the Impact of Event scale	Clusters (chi square= 10.35, df= 1, p= 0.001) Emotional representations (t= 2.56, df= 103, p= 0.012) Identity (t= 2.12, df= 115, p= 0.036)	Emotional representations (t= -2.01, df= 88, p= 0.047)
Total score	Emotional representations (r= .48, n= 55, p< 0.001) Treatment control (r= -.28, n= 58, p= 0.035)	Identity (r= .60, n= 21, p= 0.004) Emotional representations (r= .55, n= 21, p= 0.01)
Avoidance score	Emotional representations (r= .29, n= 55, p= 0.032)	Identity (r= .57, n= 21, p= 0.007) Emotional representations (r= .53, n= 21, p = 0.014).
Intrusion score	Clusters (t= 2.42, df= 49, p= 0.019) Emotional representations (r= .60, n= 56, p< 0.001) Treatment control (r= -.33, n= 60, p= 0.007) Timeline acute/chronic (r= .26, n= 59, p=0.05)	Identity (r= .51, n= 23, p= 0.014) Emotional representations (r= .45, n= 23, p= 0.032)

9.4.4 Predicting distress in the increased risk sample from illness representations

9.4.4a Predicting GHQ score[10]

A stepwise multiple regression analysis was used to predict GHQ score from the illness perception dimensions that were associated with it ($p<0.1$) (See Table 9.3). It was decided not to include the causal items in this model since there were no strong hypotheses about the contribution of causal beliefs to levels of distress in this sample. The items that were considered for entry to the model were:

- Identity
- Timeline acute/chronic
- Consequences
- Emotional representations

Table 9.9 shows the results of the analysis. Two dimensions were significant predictors of GHQ score- identity and emotional representations. Together these dimensions only accounted for 6.3% of the variance in GHQ score.

Table 9.9- Stepwise multiple regression predicting GHQ score from illness perception dimensions

Adj R square	**F**	**p**	**Significant variables**	**Std beta**	**t**	**p**
.063	6.301	0.003	Identity Emotional representations	.228 .211	2.34 2.16	0.021 0.033

When this analysis was conducted without emotional representations included as a predictor only identity was a significant predictor of GHQ score. This model accounted for 10% of the variance in general distress (see Table 9.10).

[10] Similar models were obtained using a logarithmic transformation of GHQ score

Table 9.10- Stepwise multiple regression predicting GHQ score from illness perception dimensions (except emotional representations)

Adj R square	F	p	Significant variables	Std beta	t	p
.101	12.83	0.001	Identity	.331	3.58	0.001

9.4.4b Predicting cancer worry score

A stepwise multiple regression analysis was used to predict cancer score from the illness perception dimensions that were associated with it ($p<0.1$) (see Table 9.3). The items entered into the model were:

- Identity
- Timeline acute/chronic
- Consequences
- Emotional representations
- Age (see 7.4.2a, page 207).

Table 9.11 shows the results of the analysis. Emotional representations and age were found to be a significant predictors of cancer worry accounting for 45% of the variance in cancer worry score. [11]

Table 9.11- Stepwise multiple regression predicting cancer worry score from illness perception dimensions

Adj R square	F	p	Significant variables	Std beta	t	p
.454	42.53	<0.001	Emotional representations	.658	8.89	<0.001
			Age	-.192	-2.6	0.011

When this analysis was conducted without emotional representations included as a predictor two other illness perception dimensions were found to predict cancer worry score (timeline acute/chronic and identity). Age was also a significant predictor in

[11] When age was removed from the model emotional representations alone accounted for 42% of the variance in cancer worry score (std beta= .654, t=8.603, p<0.001).

this model. Together these dimensions accounted for 12% of the variance in cancer worry score (see Table 9.12).[12]

Table 9.12- Stepwise multiple regression predicting cancer worry score from illness perception dimensions (except emotional representations)

Adj R square	F	p	Significant variables	Std beta	t	p
.119	5.75	0.001	Timeline acute/chronic	.230	2.47	0.015
			Identity	.209	2.25	0.026
			Age	-.192	-2.11	.038

9.4.5 What are the *best* predictors of distress among women at increased risk of breast cancer?

Analysis was conducted to determine the best predictors of distress measures including both the illness perception dimensions and experience variables previously used to predict distress in the increased risk sample (see section 7.4.5, page 213 and section 9.4.4 page 271)

9.4.5a What are the best predictors of general distress?[13]

A stepwise multiple regression analysis was used to predict GHQ score from the experience items and IPQ-R subscale that were associated with it ($p<0.1$). The items entered into the model were:

Experience items:

- Recency of bereavement of a personally known relative from breast cancer (plus dichotomous variable categorising if participants had or had not experienced a recent bereavement)
- If the index relative was the participants mother or another relative (dichotomous variable)

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[12] When age was removed from this model Timeline acute/chronic and Identity subscales accounted for 9% of the variance in Cancer Worry Scores ($F= 6.21$, $p= 0.003$).

[13] Similar models were obtained using a logarithmic transformation of GHQ score

- Subjective experience item 12: '*To what extent have your experiences been positive?*'

IPQ-R subscales:

- Identity
- Timeline acute/chronic
- Consequences
- Emotional representations

The results of the regression model are provided in Table 9.13. The results indicate that two of the IPQ-R subscales (identity and emotional representations) were significant predictors of distress in this sample ($p<0.05$). Together these variables account for 11% of the variance in GHQ score ($p<0.01$). Recency of bereavement did not achieve significance ($t=1.93$, $p= 0.056$).

Table 9.13- Stepwise multiple regression predicting GHQ score from experience items and IPQ-R subscales.

Adj R square	**F**	**p**	**Significant variables**	**Std beta**	**t**	**p**
.108	6.74	0.002	Identity	.233	2.36	0.021
			Emotional representations	.226	2.28	0.025

When the analysis was rerun omitting emotional representations as a predictor the revised model accounted for a slightly larger proportion of the variance in GHQ score (14%). The significant variables in this model were: identity ($p<0.01$) and recency of bereavement ($p<0.05$). The extent to which participants reported their experience had been positive did not achieve significance ($t=1.86$, $p= 0.066$).

Table 9.14- Stepwise multiple regression predicting GHQ score from experience items and IPQ-R subscales (except emotional representations).

Adj R square	F	p	Significant variables	Std beta	t	p
.14	8.85	<0.001	Identity	.286	3.01	0.003
			Recency of bereavement	-.215	-2.27	0.026

9.4.5b What are the best predictors of cancer worry score?

A stepwise multiple regression analysis was used to predict cancer score from the experience variables and IPQ-R subscales that were associated with it ($p<0.1$) (see section 7.4.5, page 213 and section 9.4.4 page 271). The experience items entered into the model were:

- Participants age when their index relative was diagnosed with breast cancer.
- Subjective experience item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'*
- Subjective experience item 9: *'Do you feel that your role in the family has changed because of your experiences of breast cancer?'*
- Subjective experience subscale: 'Traumatic experience'.

The IPQ-R subscales entered into the model were:

- Identity
- Timeline acute/chronic,
- Consequences
- Emotional representations

Age was also included as a predictor (see 7.4.2a, page 207).

Table 9.15 indicates the regression model and the significant predictors. The model produced accounted for 43% of the variance in cancer worry. The emotional representations subscale was a very strong predictor in the model ($p<0.001$). The

experience item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'* was also a significant predictor ($p<0.05$).

Table 9.15- Stepwise multiple regression predicting cancer worry score from experience items and IPQ-R subscales

Adj R square	**F**	**p**	**Significant variables**	**Std beta**	**t**	**p**
.43	36.44	.000	Emotional representations	.601	7.54	<0.001
			Change in life plans	.183	2.30	0.024

When the analysis was rerun omitting emotional representations from the independent variables the model accounted for 18.4% in the variance of cancer worry (see Table 9.16). Two experience variables (Item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'*($p<0.01$) and participants age when their index relative was diagnosed with breast cancer($p<0.05$)) were found to be significant predictors. The identity subscale of the IPQ-R was also a significant predictor of cancer worry score in this model ($p<0.05$).

Table 9.16- Stepwise multiple regression predicting cancer worry score from experience items and IPQ-R subscales (except emotional representations)

Adj R square	**F**	**p**	**Significant variables**	**Std beta**	**t**	**p**
.18	8.49	<0.001	Experience Item 10. (Change in life plans)	.307	3.37	0.001
			Identity	.233	2.57	0.012
			Participants age at index relatives diagnosis	-.192	-2.12	0.037

9.5 SUMMARY AND DISCUSSION

9.5.1 Associations between illness perceptions and general and cancer specific distress in each sample

9.5.1a Increased risk sample (A)

Overall negative representations of breast cancer as indicated by the illness representations clusters were associated with heightened cancer worry and intrusive thoughts about breast cancer as assessed by the Impact of Event scale. A number of predicted associations between illness perception dimensions and distress were identified. In the increased risk sample a stronger illness identity and emotional representations of the disease as well as perceptions of the disease as long lasting and holding greater consequences were associated with higher levels of both general distress and cancer worry. This replicates associations found between illness perceptions and emotional response to illness in patient samples, including breast cancer patients (reviewed in Chapter 3, see 3.4.5c and 3.4.6, pages 88-90).

The intrusion and avoidance subscales of the Impact of Event scale were associated with different illness perceptions. The avoidance measure was only associated with the emotional representations subscale of the IPQ-R and none of the cognitive representations. The intrusion subscale was the only distress measure associated with representations of control. Weaker belief in the ability of treatment to control breast cancer was associated with greater intrusive thoughts about breast cancer although perception of personal control over the disease was not associated with any of the distress measures in the increased risk sample. This was surprising since research in patient populations has indicated that beliefs concerning the control and cure of disease are positively associated with adaptation (eg Heijmans and de Ridder 1998, Scharloo et al. 1998, Hagger and Orbell 2001). In addition, negative associations between beliefs over control of breast cancer and distress have also been shown in breast cancer patients (Taylor et al 1984, Buick 1997) and were expected in this sample. The control subscales of the IPQ-R, assess perceptions of control over the prognosis and recovery of disease. Although these perceptions are of importance for patient samples it is likely that beliefs about the control of *risk* and *prevention* are

more important for individuals at risk of disease. In this context beliefs concerning efficacy of detection methods (BSE, mammography) and personal control over breast cancer risk (ie the impact of health related behaviours on risk) may be more crucial. Indeed women at increased risk of breast cancer who believed more strongly in a number of uncontrollable causal items (including altered immunity, poor medical care in the past, accident or injury) showed higher levels of general distress and reported greater avoidance of breast cancer as assessed by the Impact of Event scale. An in-depth discussion on perceptions of control of risk and prevention of breast cancer in relation to this work is provided in Chapter 11 (see 11.2, page 313).

The intrusion subscale of the Impact of Event scale was associated with different causal items than the avoidance subscale. The intrusion subscale was associated with internal causal items (e.g. believing breast cancer to be caused by patients behaviour or personality). Previous research has indicated that health related intrusive thoughts in healthy individuals might be associated with feelings of personal responsibility for prevention (Freeston et al. 1994). In this sample it is possible that perceiving personal characteristics as causes of breast cancer may prompt intrusive thoughts about the disease.

Stronger associations between illness representations and cancer specific distress compared to general distress were expected in the increased risk sample. Emotional representations showed much stronger correlations with cancer specific distress measures than general distress. The high correlations between emotional representations and cancer specific distress measures in all samples suggested that there was a degree of conceptual overlap between these constructs. In the increased risk sample the strength of association between cognitive representations and distress measures were similar for both general and cancer specific distress. This suggests that illness representations are equally associated with general distress and anxiety specifically concerning breast cancer.

9.5.1b Control sample (C)

As predicted, few associations between illness representations and distress were identified in the control sample of women without any experience of breast cancer.

General distress was only associated with emotional representations. The only cognitive representation associated with any measure of distress in this sample was the identity dimension. This is consistent with the SRM that proposes that feelings of vulnerability are an important prerequisite for linking illness representations with emotional response. Representations of breast cancer in the control sample are unlikely to be considered personally relevant or threatening and therefore do not provoke anxiety.

9.5.2 Predicting levels of general and cancer specific distress in the increased risk sample

Multiple regression analyses indicated that dimensions of illness representations were significant predictors of general distress and cancer specific distress. Women's emotional representation of breast cancer and perceptions of the symptoms associated with the disease (identity) were predictors of both general distress and cancer worry. When assessing the predictive power of the *cognitive* representations identity was the best predictor of general distress. Cancer worry was predicted by identity and perception of the duration of breast cancer. Age was also found to be an independent predictor in this model (younger age predicting higher levels of cancer worry).

Analysis was conducted in order to determine the best predictor of distress in this sample from the experience, illness perception and demographic variables. This was a clinically driven research question in order to identify predictors of distress that may be recognised by genetic counsellor and staff at the risk clinics as well to highlight possible predictors that may be amenable to therapeutic intervention. When the experience items and illness representations were entered into the analysis together both were found to predict general distress and cancer worry. The measure of emotional representations was found to be a strong predictor of both general and cancer specific distress and appeared to mask a number of other predictor variables. It was important to omit this dimension from the analysis in order to examine the predictive value of those masked variables. Both general distress and cancer worry were found to be best predicted by women's perception of the identity of breast cancer and aspects of experience of the disease in the family. However, different

types of experience were found to predict either general distress or cancer worry with the identity dimension. Identity and recency of bereavement were found to be the best predictors of general distress. For cancer worry identity, changes in life plans and age of the participant when their index relative was diagnosed were found to be the best predictors. This supports the hypothesis previously outlined in Chapter 4 that general distress would be associated with issues of bereavement whereas cancer specific worry was predicted to be associated experiences directly influencing the life and future of the individual at risk.

Although a number of variables were indicated as important predictors of distress in this sample the proportion of variance explained by both the experience variables and illness perception measures were disappointing. It is possible that other unmeasured variables may be contributing to levels of distress (eg concern over passing the genetic predisposition to ones children). It is also possible that the measures used require additional work to improve validity (see Chapter 6). At this stage conclusions can not be made concerning explanation of distress or causal relations between variables in this analysis. Regression analysis is based on correlations and no firm conclusions can be made regarding the causality of variables in this analysis (Cohen and Cohen 1983). Alternative explanations regarding causality will be discussed in more detail in the following chapter (see section 10.5.3, page 304) and attempts to further explore issues of causality are discussed in Chapter 11 (see 11.3.2, page 320).

CHAPTER 10

TESTING THE MEDIATION MODEL

TESTING THE MEDIATION MODEL

10.1 AIMS AND RATIONALE

The previous chapters in this thesis have demonstrated associations between: experience of breast cancer and distress (Chapter 7); experience of breast cancer and illness perceptions (Chapter 8); illness perceptions and levels of distress (Chapter 9). These sets of analyses were conducted in order to explore systematically the mediation model outlined in Chapter 4 (see Figure 4.1, page 100). This model was developed from the Self Regulatory Model, which proposes that response to health threats are determined by an individual's cognitive and emotional representation of the threat derived from their experiences (Leventhal et al. 1980). The previous chapters confirmed that the three sets of variables (experience, illness perceptions and distress) were interrelated and that both aspects of experience and dimensions of illness representations predicted levels of general and cancer specific distress in women at increased risk of breast cancer. However, this analysis does not offer any explanation for the causal mechanisms involved. The analysis outlined in this Chapter was conducted in order to address the primary aim of the research and test the mediation model. It was hypothesised (H8) that the impact of experience of breast cancer on levels of general and cancer specific distress will be mediated by perceptions of breast cancer.

10.2 MEASURES AND SAMPLES

The design and procedure were previously provided in Chapter 4. The samples reported in this study included: Increased risk sample and control sample. These samples were described in Chapter 5.

The measures utilised in this analysis were:

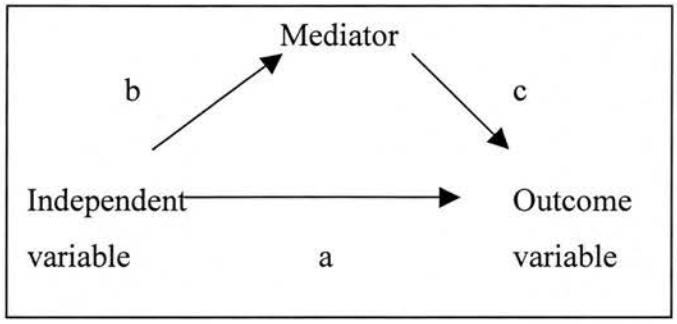
- Experience questionnaire (increased risk sample only) (see Chapter 6, Part 2, section 6.3-6.8)
- General distress (GHQ-30) (see Chapter 4, section 4.4.4a, page 106)
- Cancer specific distress (Cancer worry scale, Impact of Event scale) (see Chapter 4, section 4.4.4b, page 107)
- IPQ-R adapted to assess healthy women's perceptions of breast cancer (see Chapter 6, Part 3, section 6.9-6.15)

10.3 STATISTICAL METHODS

10.3.1 Mediation

Mediation refers to the process by which an independent variable is thought to influence the dependent variable (Baron and Kenny 1986). Mediating variables are often internal psychological variables that explain the links between stimuli and behavioural outcome. A mediator is identified if it is able to account for the associations between the independent and dependent variable. Baron and Kenny (1986) illustrate the process of mediation in the following model:

Figure10.1- An illustration of mediation



When there is a significant association between the independent variable and the outcome variable (a) another variable may be considered to mediate this association. Mediation is confirmed is the following conditions are met:

- Variations in the independent variable significantly account for variations of the mediator (b)
- Variations in the mediator significantly account for variations in the outcome variable (c)
- When the pathway via the mediator is controlled (b-c) the association between the independent and outcome variable (a) becomes non significant.

Perfect mediation is said to occur if the association between the independent variable and dependent variable is eliminated entirely when the mediation pathway is controlled. However, many psychological constructs such as distress have multiple causal factors. A *reduction* in the association between the independent and outcome

variable is therefore taken to represent mediation in this field (Baron and Kenny 1986).

Baron and Kenny (1986) refer to a 'approximate significance test' for mediation. This is known as the 'Sobel Test' (Sobel 1982). This tests that the indirect effect of the IV on the DV via the mediator is significantly different from zero.

The formula for this test is:

$a*b/SQRT\ (b^2*s_a^2 + a^2*s_b^2)$

Where

a	Raw (unstandardized) regression coefficient for the association between the IV and mediator
s_a	Standard error of *a*
b	Raw regression coefficient for the association between the mediator and the DV (when the IV is also a predictor of the DV)
s_b	Standard error of *b*

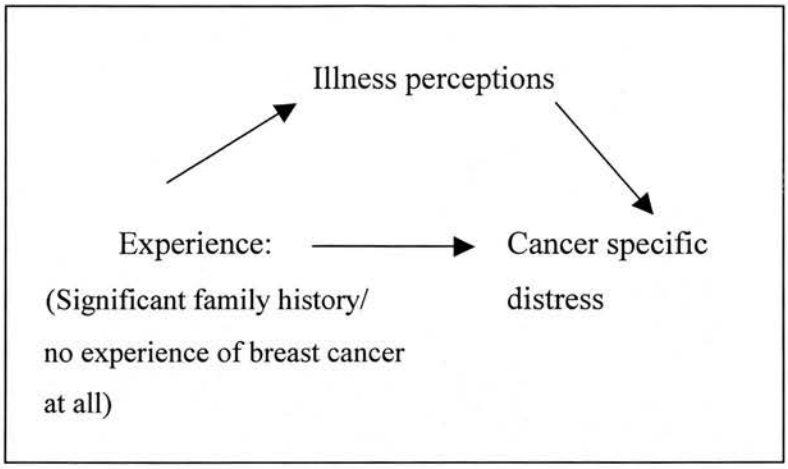
An interactive website is available to calculate this test: http://quantrm2.psy.ohio-state.edu/kris/sobel/sobel.htm (Preacher and Leonardelli 2001, accessed March 2003).

This test was calculated for all models that suggested mediation. The Sobel test statistic and p value are reported. Full details of calculations are reported in the Appendix III, (A-24)

10.3.2 Potential mediation models

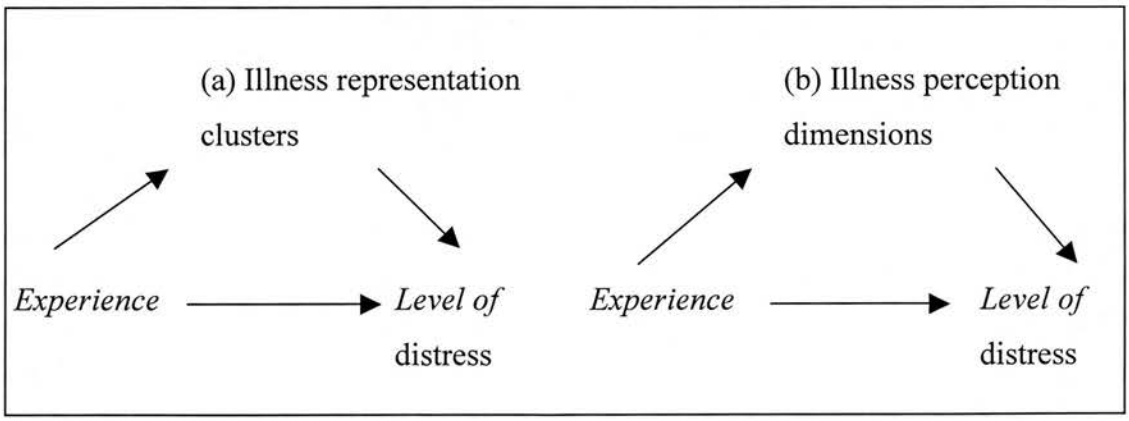
The mediation model proposed in this thesis could be examined at different levels. Firstly it was predicted that the difference in levels of cancer specific distress between the samples with different experiences of breast cancer (women at increased risk and the control sample of women with no experience of breast cancer) would be mediated by differences in illness perceptions (See Figure 10.2).

Figure 10.2- Differences in levels of cancer specific distress between samples with different experience of breast cancer: Mediation via illness perceptions



Secondly, it was predicted that associations between experience of breast cancer in the family and levels of distress in the increased risk sample would be mediated by illness perceptions. Mediation via the overall pattern of illness perceptions as represented by the clusters (see Figure 10.3(a)) and specific subscales (see Figure 10.3 (b)) were examined.

Figures 10.3(a-b)- Associations between experience of breast cancer in the family and levels of distress: Mediation via illness representation clusters (a) and illness perception dimensions (b)



10.3.3 Steps of analysis

Step 1. Identify independent variables

Associations between experience of breast cancer and continuous distress measures suitable for multiple regression (GHQ score and cancer worry score) were examined in order to identify independent variables for potential mediation models. Significant correlations between experience items and distress measures have previously been reported (Chapter 7) and simple regression models were used to determine the predictive value of the independent experience variable on the distress outcome measures.

Step 2. Identify potential mediators

For each independent experience variable found to be significant in predicting a distress outcome measure potential mediators were identified. Potential mediators were either illness representation clusters or illness perception dimensions that had previously been identified as being significantly associated with the independent experience variable (Chapter 8). Logistic regression was used to determine the predictive value of the independent variable on categorical mediator variables (illness representation clusters) (see 10.3.4, page 287) and simple linear regression was used to determine the predictive value of the independent variable on continuous mediators variables (illness perception dimensions).

Step 3. Test the mediation model

The procedure outlined by Baron and Kenny (1986) was used to test the mediation models identified. The potential mediator and independent variable were entered consecutively into a multiple regression model. Firstly checks were made to determine if the mediating variable was a significant predictor of the independent variable. If so, the effect of the independent variable on the dependent variable in this model was compared to the effect of the independent variable on the dependent variable alone. If the effect of the independent variable was reduced in the final model mediation was considered to have occurred and the Sobel test conducted (Sobel 1982, Preacher and Leonardelli 2001). Simultaneous (standard) multiple regression was used because it allows the researcher to specify the variables to include in the model (see Chapter 8 for details of multiple regression techniques).

Where alternative mediation models are found (i.e. more than 1 mediator is found to mediate the effect of an independent variable on the same dependent variable) a third model is tested including all potential mediators in the same model.

10.3.4 Logistic regression

Logistic regression was used to check the predictive value of experience variables on categorical mediators (ie illness representation clusters). Logistic regression has similar aims to multiple regression but in this case the dependent variable is categorical rather than continuous. This makes the data unsuitable for multiple regression because the assumptions concerning univariate and multivariate distribution will not hold with a dichotomous dependent variable.

In logistic regression the dichotomous dependent variable is transformed using a logistic transformation. The transformed variable can then be predicted using logistic regression in a manner analogous to linear regression models. The aim of logistic regression is to find the best linear combination of independent variables to maximise the likelihood of achieving the observed category membership. The coefficients in logistic regression are measures of the changes in odds ratio (ratio of probabilities). As in multiple regression the analysis can be used to identify predictors, assess the importance of predictors and assess strength of association.

Predictor variables may be continuous, dichotomous or a mixture and no assumptions are made about the distribution or homogeneity of predictor variables. However the technique is still sensitive to the problems of multicollinearity and sample size. The techniques for selecting variables are similar to that outlined in multiple regression and include standard, hierarchical and statistical regression.

The regression coefficients are interpreted the same as in multiple regression. Standardized coefficients are tested to determine if they are significantly different from zero. Exponentials of the coefficients can also be obtained which show the effects of the independent variable on the odds of the dependent variable.

The statistics examined from this analysis were:

- Model Chi-square (Tests if the log likelihood of the model is significant compared to the constant only model).
- Wald statistic (Indicates if the predictor is reliably associated with outcome)
- Standardized regression coefficients
- Exponential regression coefficients (Change in the odds of being in one group of the dependent variable produced by a change in one unit in the independent variable).
- Sobel test (Tests that the indirect effect of the IV on the DV via the mediator is significantly different from zero, see 10.3.1).

10.4 RESULTS

10.4.1 Differences in levels of cancer specificdistress between the increased risk and control sample: Mediation via illness perceptions.

Dependent variables

Tests for differences between the sample of women at increased risk of breast cancer (Sample A) and the control sample (Sample C) were previously examined and differences in levels of cancer worry were identified (see chapter 7). When 'sample' was coded as a binary variable regression analysis confirmed that 'sample' significantly predicted cancer worry score (Adjusted R square= .12, f= 30.95, $p< 0.001$, standardized beta= .356, t= 5.56, $p< 0.001$).

Mediators

'Sample' was found to be a significant predictor of three of the illness perception dimensions: illness coherence, emotional representations and consequences ($p<0.05$). The predictive value of the binary variable 'sample' on each of these potential mediators is provided in Table 10.1.

Table 10.1- Illness perception dimensions significantly predicted by 'sample'.

IPQ-R subscales predicted by 'sample'	**Adj. R Square**	**F**	**p**	**Std. Beta**	**t**	**p**
Illness coherence	.18	44.19	<0.001	-.423	-6.65	<0.001
Emotional representations	.018	4.65	0.032	.153	2.16	0.032
Consequences	.014	3.89	0.014	.138	1.97	0.05

Potential mediation models

Three potential mediation models were tested. These are shown diagrammatically below (Models 10.1(i)-(iii)). Each model was tested separately and the results of the regression analysis are provided in Table 10.2.

Models 10.1(i)-(iii) Effect of 'sample' on cancer worry mediated by illness perceptions

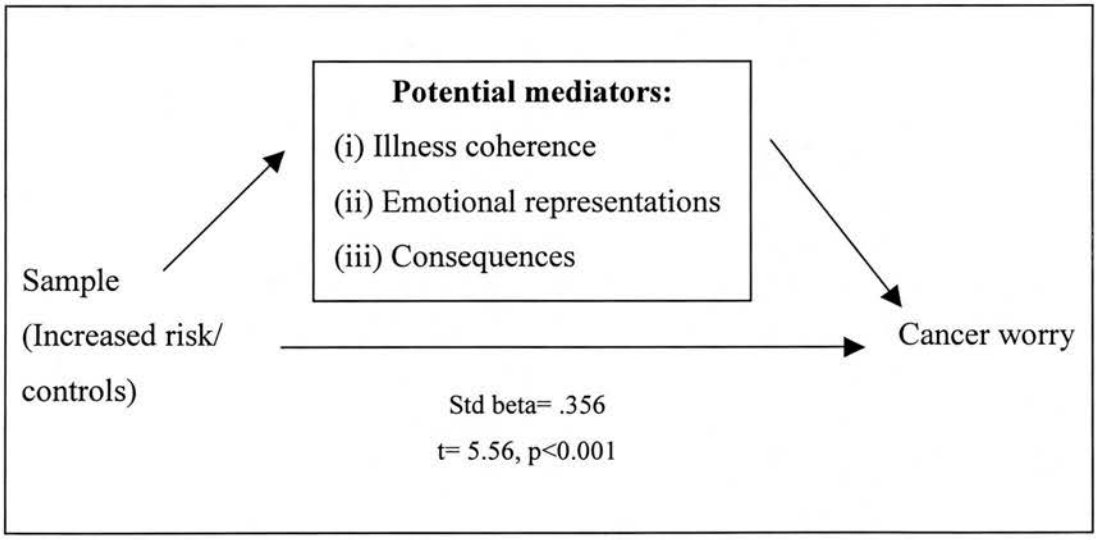


Table 10.2- Testing three mediation models of illness perceptions mediating the association between 'sample' and cancer worry

Model	Adj. R Square	F	p		Std. Beta	t	p
10.1(i)	.118	14.50	<0.001	**Mediator**	.051	.70	0.49
				IV	.374	5.15	< 0.001
10.1(ii)	.426	72.64	<0.001	**Mediator**	.562	10.17	<0.001
				IV	.262	4.75	<0.001
10.1(iii)	.141	17.53	<0.001	**Mediator**	.176	2.66	0.008
				IV	.321	4.86	<0.001

Model 10.1(i)

The results indicated that illness coherence was not a mediator, as it did not significantly predict cancer worry in the model ($p<0.05$).

Model 10.1(ii)

Both 'sample' and emotional representations were significant predictors of cancer worry in the model ($p<0.05$). The predictive value of 'sample' when predicting cancer worry alone (standardized beta= .356, t= 5.56) was higher than compared to when 'sample' was entered with emotional representations (standardized beta= .262, t= 4.75). Including the mediator in the model improves the fit of the model and reduced the magnitude of the coefficient of the independent variable. This suggests that the mediator is partially replacing the independent variable as a predictor of the dependent variable (cancer worry). The sobel test indicated that the mediator was significant (Sobel test statistic= 2.106, p= 0.035).

Model 10.1(iii)

Both 'sample' and consequences were significant predictors of cancer worry in the model ($p<0.05$). The effect of sample was lower when entered with consequences (standardized beta= .321, t= 4.86) compared to when it is in the model alone (standardized beta= .356, t= 5.56). This suggests that perceptions of the consequences of breast cancer partially mediate the effect of sample on levels of cancer worry. However, the sobel test indicated that the mediator was not significant (Sobel test statistic= 1.581, p=0.114).

Combined models

Since both emotional representations and consequences were found to mediate the same effect (see model 10.1(ii) and 10.1 (iii)) another model was tested to include both of these mediators. The results of this analysis are provided in Table 1.3. The results indicated that when assessed together the emotional representation subscale is a mediator of the effect of sample on cancer worry whereas the consequences subscale is not.

Table 10.3- Testing the combined mediation model that emotional representations and consequences mediate the association between 'sample' and cancer worry

Model	**Adj.R Square**	**F**	**p**		**Std. Beta**	**t**	**p**
Sample + emotional representation + consequences predicting cancer worry score	.396	73.61	<0.001	**Emotional representations**	.568	12.67	<0.001
					-.015	-.349	.727
				Consequences IV	.217	5.003	<0.001

10.4.2 Mediation models in the increased risk sample

10.4.2a General distress

Independent variables

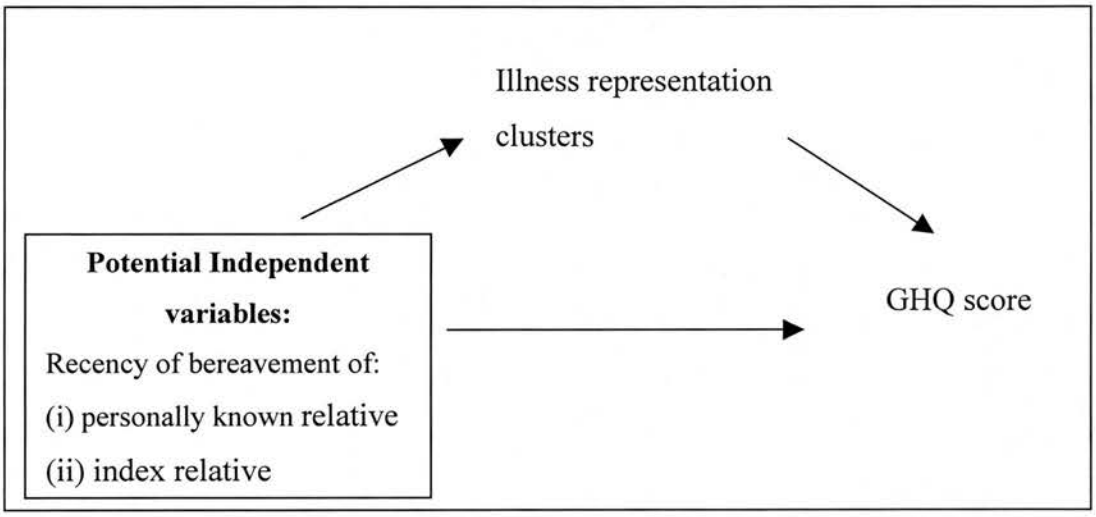
Correlational analysis reported in chapter 7 revealed that GHQ score was significantly correlated with recency of bereavement of both a personally known relative and the index relative from breast cancer ($p<0.05$). Simple regression analysis confirmed this result:

- Recency of bereavement of a personally known relative (Adjusted R square= .086, f= 8.91, p= 0.004, standardized beta = -.311, t= -2.90, p= 0.004)
- Recency of bereavement of index relative (Adjusted R square= .052, f= 4.79, p= 0.032, standardized beta= -.257, t= -2.19, p= 0.032).

Clusters as mediators

The results reported in Chapter 8 indicated that the individuals in each of the illness representation clusters differed on the recency of bereavement variables. Logistic regression confirmed that these experiences variables were significant predictors of the binary illness representations cluster variable. (Recency of bereavement of a personally known relative: chi-square= 8.18, df= 1, p= 0.004, Exp beta= 1.058, p= 0.007; Recency of bereavement of index relative: chi-square= 5.48, df= 1, p= 0.019, exp beta= 1.005, p= 0.024). This suggested two possible mediation models (Models 10.2 (i) and (ii))

Models 10.2 (i-ii)- Effect of recency of bereavement on GHQ score mediated by illness representation clusters.

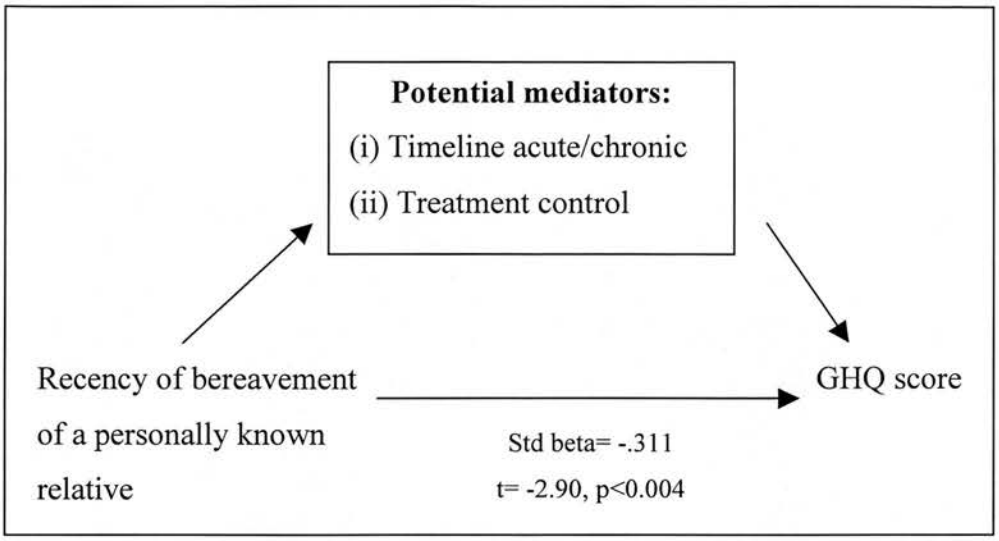


In models 10.2 (i) and 10.2 (ii) illness representation cluster variable was not found to be a significant predictor of GHQ score ($p<0.05$) and therefore the model did not meet the criteria for mediation.

Illness perception dimension as mediators

Associations between experience items and illness perceptions were outlined in chapter 8. Recency of bereavement was predicted by the IPQ-R subscales timeline acute/chronic (Adjusted R square= .082, f= 6.998, p= 0.01, standardized beta= -.287, t= –2.65, p= 0.01) and treatment control (Adjusted R square= .082, f= 7.10, p= 0.009, standardized beta= .287, t= 2.67, p= 0.009). This suggested two possible mediation models:

Models 10.3(i-ii) Effect of recency of bereavement mediated by illness perceptions



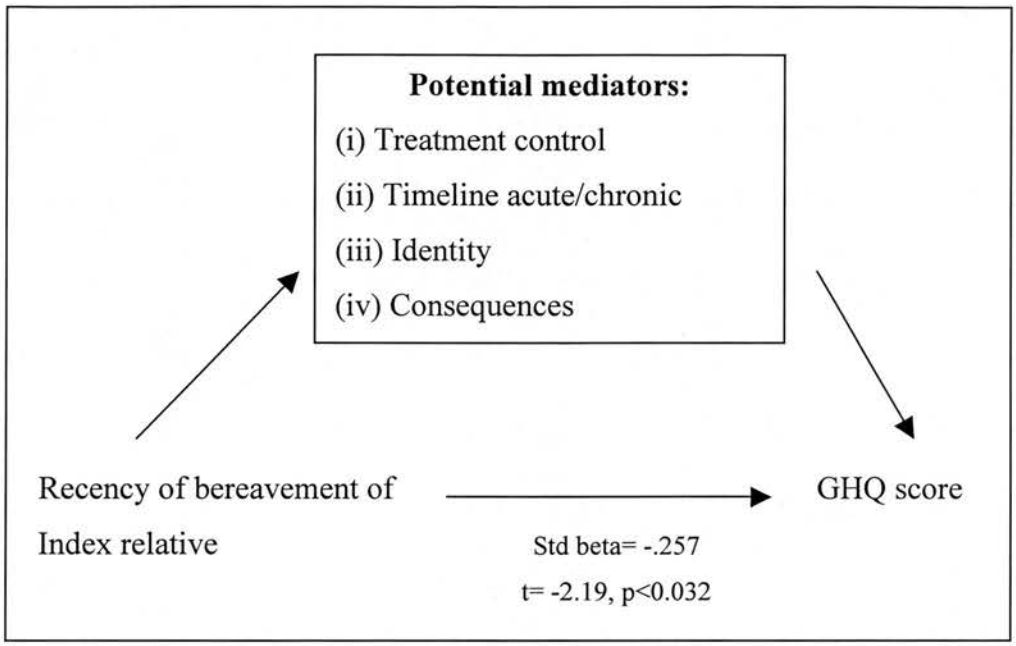
In models 10.3 (i) and 10.3 (ii) the IPQ-R subscales timeline acute/chronic and treatment control were not significant predictors of GHQ score ($p<0.05$). Therefore there was no mediation through these variables.

Recency of bereavement of the index relative from breast cancer predicted a number of illness perception dimensions:

- Treatment control (Adjusted R square= .102, f= 8.48, p= 0.005, standardized beta .340, t= 2.91, p= 0.005);
- Timeline acute/chronic (Adjusted R square= - .088, f= 7.25, p= 0.009, standardized beta= -.319, t= -2.69, p= 0.009);
- Identity (Adjusted R square= .059, f= 5.43, p= 0.023, standardized beta= -.270, t= –2.33, p= 0.023);
- Consequences (Adjusted R square = .052, f = 4.54, p= 0.037, standardized beta= -.257, t=-2.13, p= 0.037)

The following mediation models were therefore tested:

Models 10.4 (i-iv)- Effect of recency of bereavement of index relative on GHQ score mediated by illness perceptions



In models 10.4 (v) and 10.4 (ii) the illness perceptions dimensions treatment control and timeline acute/chronic were not significant predictors of GHQ score ($p<0.05$). Therefore the model did not meet the criteria for mediation.

Table 10.4 shows that in model 10.4 (iii) identity was a significant predictor of GHQ score. When both the independent variable and mediator are entered into the model together the effect of the independent variable (recency of bereavement of index relative) became non significant ($p<0.05$). The results suggest that the effect of recency of bereavement of index relative was mediated by its effect on perceptions of the identity of breast cancer. However, the sobel test indicated that the mediator was not significant (Sobel test statistic = 1.646, $p=0.0099$)

Table 10.4- Testing the mediation model that 'identity' mediates the association between recency of bereavement of the index relative and general distress

Model 10.4 (iii)	**Adj. R Square**	**F**	**P**		**Std. Beta**	**t**	**p**
Recency of bereavement of index relative + identity predicting GHQ score	.095	4.62	0.013	**Mediator IV**	.244 -.191	2.05 -1.61	0.044 0.113

Table 10.5 shows that in model 10.4 (iv) consequences was a predictor of GHQ score (p= 0.057) and the effect of recency of bereavement of index relative became non significant (standardized beta= -.178, t=-1.14, p=0.16). This suggests that the effect of recency of bereavement of index relative on levels of general distress was also marginally mediated by its impact on beliefs about the consequences of breast cancer. However, the sobel test indicated that the mediator was not significant (Sobel test statistic= 1.940, p= 0.0524).

Table 10.5- Testing mediation model that 'consequences' mediates the association between recency of bereavement of the index relative and general distress.

Model 10.4 (iv)	**Adj. R Square**	**F**	**p**		**Std. Beta**	**t**	**p**
Recency of bereavement of index relative + consequences predicting GHQ score	.085	3.96	0.007	**Mediator IV**	.241 -.178	1.94 -1.14	0.057 0.16

Combined model

Since both identity and consequences were found to mediate the same effect (see model 10.4 (iii) and 10.4 (iv)) another model was tested to include both of these mediators. The results of this analysis are provided in Table 10.6. The results indicated that when the mediators are assessed together the identity subscale is a

significant mediator of the effect of recency of bereavement on general distress whereas the consequences subscale is not ($p<0.05$).

Table 10.6- Testing the combined mediation model that identity and consequences mediate the association between recency of bereavement and general distress

Model	**Adj.R Square**	**F**	**p**		**Std. Beta**	**t**	**p**
Recency of bereavement of index relative + identity + consequences predicting GHQ score	.137	4.39	<0.001	**Identity**	.273	2.18	.033
				Consequences	.185	1.50	.138
				IV	-.108	-.86	.393

10.4.2b Cancer worry

Independent variables.

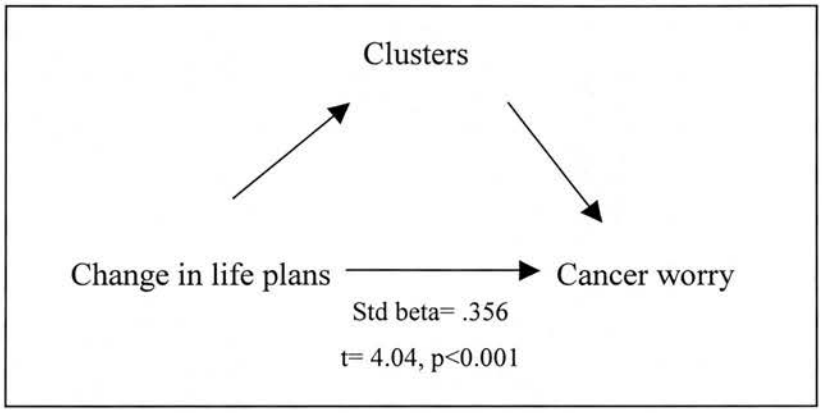
Associations between experience items and cancer worry were reported in Chapter 7 (8.4.2b(i)). Cancer worry was significantly predicted by:

- Subjective experience item 10: *'How much do you feel your life plans have changed because of the risk of cancer in your family?'* (Adjusted R square= .119. f= 16.30, p<0.001, standardized beta= .356, t= 4.04, p< 0.001)
- Subjective experience subscale: 'Traumatic experience' (Adjusted R square= .04, f= 5.49, p= 0.021, standardized beta= .221, t= 2.34, p= 0.021)
- Subjective experience item 9: *'Do you feel that your role in the family has changed because of your experiences of breast cancer'* (Adjusted R square= .034, f= 5.003, p= 0.027, standardized beta= .207, t= 2.24, p= 0.027)
- Participants age when index relative was diagnosed (Adjusted R square= .027, f= 4.13, p= 0.044, standardized beta= -.187, t= -2.03, p= 0.044).

Clusters as mediators

Of the above independent variables the only variable found to predict the illness representation clusters in a logistic regression analysis was the Subjective experience variable 10 (change in life plans) (Chi-square= 7.38, df= 1, p= 0.007, Exp B= .583, p= 0.01). This suggested a mediation model illustrated by Model 10.5:

Model 10.5- Effect of subjective experience variable 10 (Change in life plans). on levels of cancer worry mediated by illness representation clusters.



The test of this mediation model is summarised in Table 10.7. The analysis revealed that when entered into the same model both the independent variable (change in life plans) and the mediator (illness representation clusters) were significant predictors of cancer worry ($p \leq 0.01$) The effect of the independent variable was higher on its own (standardized beta = .356, t=4.40, p<0.001) than when entered into the model with clusters (standardized beta= .253, t= 2.64, p=0.01). This suggests partial mediation by illness representation clusters. However, the sobel test indicated that the mediator was not significant (Sobel test statistic = -1.77, p=0.0763).

Table 10.7- Testing mediation model that illness representation clusters mediates the association between subjective experience variable 10 (Change in life plans) and cancer worry

Model 10.5	**Adj. R Square**	**F**	**p**		**Std. Beta**	**t**	**p**
Change in life plans + clusters predicting cancer worry	.181	11.53	< 0.001	**Mediator IV**	-.306 .253	-3.18 2.64	0.002 0.01

Illness perception dimensions as mediators

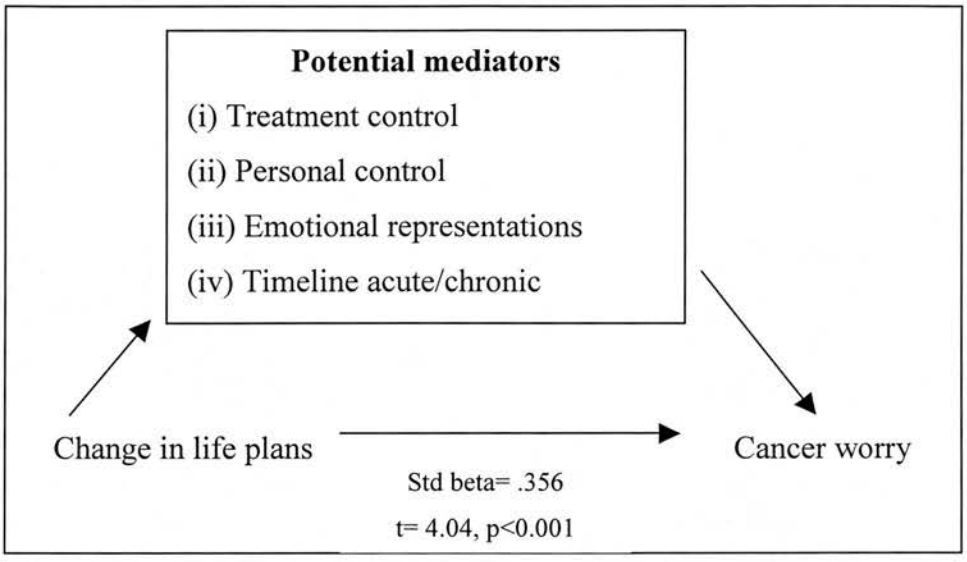
For each independent variable potential mediators were identified.

The subjective experience variable 10 (change in life plans) was found to predict the following IPQ-R dimensions:

- Treatment control (Adjusted R square= .057, f= 7.61, p= 0.007, standardized beta= -.257, t= –2.76, p= 0.007);
- Personal control (Adjusted R square= .046, f= 6.24, p= 0.014, standardized beta= -.235, t= –2.50, p= 0.014);
- Emotional representations (Adjusted R square= .038, f= 5.03, p= 0.027, standardized beta= .218, t= 2.24, p= 0.027);
- Timeline acute/chronic (Adjusted R square= .028, f= 4.11, p= 0.045, standardized beta= .192, t= 2.03, p= 0.045).

The following mediation models (Models 10.6 i-iv) were therefore tested:

Models 10.6 (i-iv)- Effect of subjective experience variable 10 (Change in life plans) on cancer worry mediated by illness perceptions.



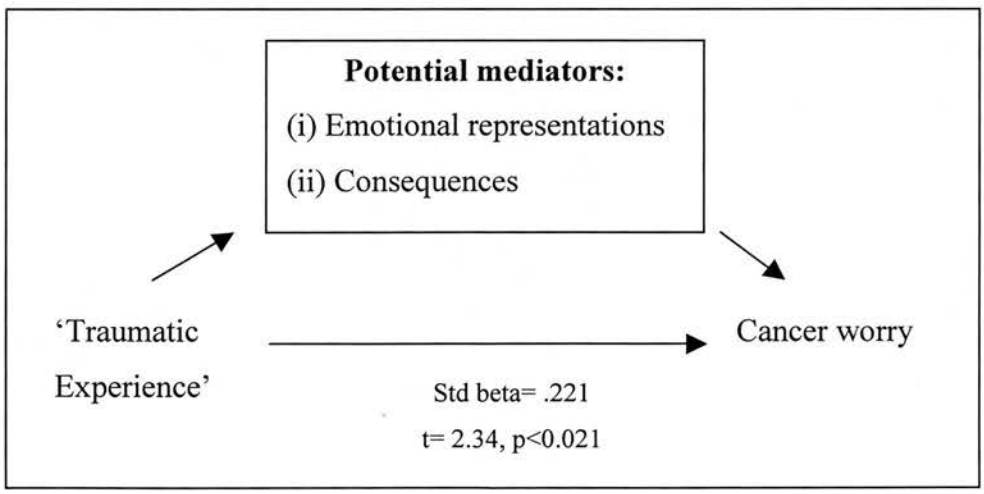
None of the cognitive representations in models 10.6 (i, ii, and iv) were found to be significant predictors of cancer worry. Only emotional representations (Model 10.6 (iii)) was a significant predictor of cancer worry ($p<0.05$). The results of this analysis are summarised in Table 10.8. Both the mediator (emotional representations) and independent variable (change in life plans) were significant predictors of cancer worry in the model. However the effect of the independent variable in this model (standardized beta= .180, t= 2.33, p= 0.022) was lower compared to when predicting cancer worry alone (standardized beta= .356, t= 4.04, $p<0.001$). This suggests partial mediation of the independent experience variable 10 (change in life plans) on cancer worry by emotional representations. The Sobel test confirmed that the mediator was significant (Sobel test statistic= 2.158, p= 0.031).

Table 10.8- Testing the mediation model that emotional representations mediates the association between subjective experience variable 10 (Change in life plans) and cancer worry

Model 10.6 (iii)	**Adj. R Square**	**F**	**P**		**Std. Beta**	**t**	**p**
Change in life plans+ emotional representations predicting cancer worry	.428	38.83	< 0.001	**Mediator** **IV**	.60 .180	7.79 2.33	< 0.001 0.022

The experience subscale 'Traumatic experience' significantly predicted two illness perception dimensions: Emotional representations (Adjusted R square= .126, f= 15.04, p< 0.001, standardized beta= .368, t= 3.88, p< 0.001); Consequences (Adjusted R square= .07, f= 8.59, p= 0.004, standardized beta= .281, t= 2.93, p= 0.004). This suggested two potential mediation models illustrated in Models 10.7 (i-ii):

Models 10.7 (i-ii)- Effect of subjective experience subscale 'Traumatic experience' on levels of cancer worry mediated by illness perceptions.



In model 10.7(ii) consequences was not found to be a significant predictor of cancer worry in the model (p<0.05) and hence no mediation was supported.

In model 10.7(i) emotional representations was found to be a significant predictor of cancer worry (p<0.05). The effect of the independent variable ('Traumatic

experience') became non significant and the beta value reduced dramatically to (standardized beta = -.039, t=-.46, p= 0.65) in this model than when predicting the cancer worry independently (standardized beta= .221, t= 2.34, p= 0.021) (see Table 10.9). This indicated that emotional representations of breast cancer mediate the impact of 'Traumatic experience' of breast cancer in the family on levels of cancer worry. The Sobel test confirmed that the mediator was significant (Sobel test statistic= 3.46, p= 0.00055).

Table 10.9- Testing the mediation model that emotional representations mediates the association between subjective experience subscale 'Traumatic experience' and cancer worry

Model 10.7 (i)	**Adj. R Square**	**F**	**p**		**Std. Beta**	**t**	**p**
Trauma+ emotional representations predicting cancer worry	.403	33.44	<0.001	**Mediator IV**	.658 -.039	7.73 -.46	<0.001 0.65

The subjective experience item 9 *('Do you feel that your role in the family has changed because of your experiences of breast cancer?')* was found to predict the IPQ-R subscale emotional representations (Adjusted R square= .032, f= 4.34, p= 0.04, standardized beta= .203, t= 2.08, p= 0.04). The following model was therefore tested (Model 10.8):

Model 10.8- Effect of subjective experience item 9 (Role change) on levels of cancer worry mediated by emotional representations.

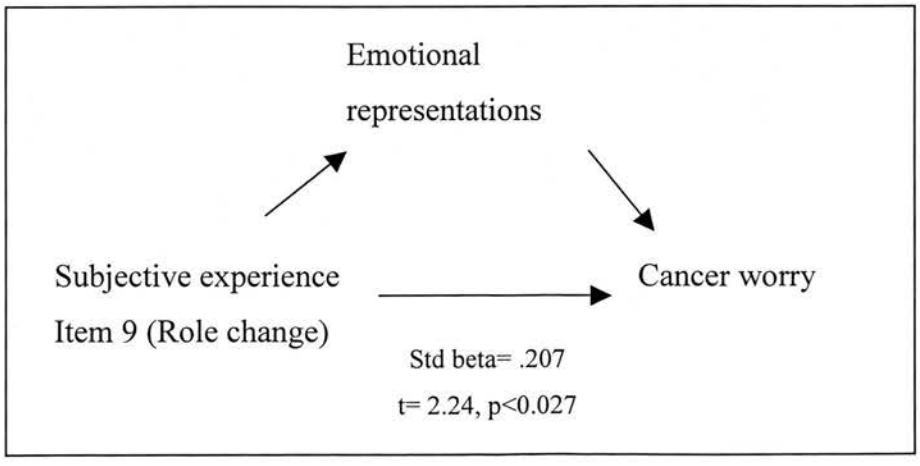


Table 10.10 provides a summary of the analysis of model 10.8. The mediator (Emotional representations) was found to be a significant predictor of cancer worry. The effect of the independent variable (role change) became non significant (standardized beta = .153, t= 1.97, p= 0.052) compared to when alone in the model (standardized beta = .207, t= 2.24, p= 0.027). This indicates that emotional representations mediates the impact of the subjective experience item 9 (Role change) on levels of cancer worry. The Sobel test confirmed that the mediator was significant (Sobel test statistic= -1.77, p= 0.0763).

Table 10.10- Testing the mediation model that emotional representations mediates the association between subjective experience item 9 (Role change) and cancer worry

Model 10.8	**Adj. R Square**	**F**	**p**		**Std. Beta**	**t**	**p**
Role change + emotional representations predicting cancer worry	.420	37.52	<0.001	**Mediator** **IV**	.608 .153	7.85 1.97	<0.001 0.052

10.5 SUMMARY AND DISCUSSION

10.5.1 Differences in levels of cancer specific distress between the increased risk and control samples: mediation by illness perceptions.

Previous analysis (Chapter 7) had indicated that although there was no difference between the increased risk sample and control sample on levels of general distress the increased risk sample showed significantly higher levels of cancer worry. The samples also differed significantly on a number of dimensions of illness perceptions including illness coherence, consequences and emotional representations (see Chapter 8). It was possible therefore that the difference in level of cancer worry between the samples may to some extent be explained by differences in their illness representations. These mediation models were tested and the results suggested that women with a significant family history of breast cancer who are at increased risk of

developing the disease have stronger emotional representations of breast cancer and perceive the disease to hold more consequences. These representations account for some of the variation in levels of cancer worry between the samples although only the emotional representations subscale was found to be a significant mediator. In addition, sample membership was still a strong predictor of cancer worry suggesting that there are other factors that differ between the samples (for example risk status, risk perception etc) that may also contribute to this effect.

10.5.2 Mediation models in the increased risk sample

In the increased risk sample recency of bereavement was the only experience variable that significantly predicted levels of general distress. General distress was also associated with the overall pattern of beliefs held by participants (illness representation clusters) and a number of dimensions of illness perceptions (identity, consequences, timeline acute/chronic and treatment control). The illness representation clusters, perception of the duration of breast cancer (timeline acute/chronic) and beliefs concerning the ability of treatment to control the disease (treatment control) were not significant predictors of GHQ score and could not act as mediators. Recency of bereavement of the index relative was found to be mediated by perceptions of the identity of breast cancer and consequences of the disease although these effects were not shown to be significant. Women who had lost their index relative more recently perceived breast cancer to hold more severe consequences and believed more symptoms to be associated with breast cancer. The identity dimension was found to be the strongest mediator in a joint model. These results suggested that the effect of bereavement on levels of distress in women at increased risk of breast cancer might not only reflect issues concerning grief but also the impact of the experience on women's perceptions of breast cancer and subsequently the meaning of their own risk. Previous research suggested that genetic counselling provokes a reactivation of grief (DudokdeWit et al. 1997, Lodder et al. 1999, Hopwood et al. 1998). This reactivation may not only concern the sense of loss but also invoke representations of breast cancer. The more recent the bereavement the more accessible and threatening these images may be (see 3.2.1a, page 73).

Cancer worry was associated with a number of experience variables and illness perception subscales suggesting a number of possible mediation models. Illness representation clusters were found to mediate the association between the experience variable *'How much do you feel your life plans have changed because of the risk of cancer in your family?'* and cancer worry although this effect did not reach significance. This suggested that women who reported greater changes in their life plans because of the risk of breast cancer in their family had more negative representations of the disease which led to increased cancer worry. When individual dimensions of illness perceptions were examined none of the cognitive representations were found to be mediate this effect. The emotional representations subscale was the only dimension found to contribute to the effect. Emotional representations appeared to be a significant mediator of experience of breast cancer in the family on cancer worry. A number of aspects of experience assessed in this study (including how traumatic the experience had been, changes in life plans because of breast cancer risk and changes in family roles due to breast cancer) were found to be mediated by emotional representations of breast cancer. This suggests that it is the fear aroused by these experiences of breast cancer that promotes cancer worry.

The number of mediation models found in the data was disappointing. None of the cognitive representations were found to significantly mediate experiences of breast cancer in the family on levels of cancer specific distress. A number of methodological problems may have led to difficulties in detecting mediation effects.

10.5.3 Methodological issues

Baron and Kenny (1986) discuss a number of assumptions of using this technique to test mediation models. One of the main assumptions is that the dependent variable does not cause the mediator. In the analysis reported in this chapter there were strong theoretical reasons for hypothesising that the mediator (illness representations) caused the dependent variable (level of distress). However it is also possible that distress may influence illness representations. Measures of distress are likely to encompass anxiety related dispositions such as negative affectivity. Negative affectivity is a dispositional dimension that reflects individual differences in negative

emotionality (Watson and Clark 1984). Individuals showing high negative affectivity tend to be more distressed, upset and perceive a negative view of themselves and the world. The construct overlaps with neuroticism and trait anxiety and reflects both affective states and the tendency to experience negative emotions as well as styles of perceiving, recalling and reporting events (Costa and McCrae 1985, Watson and Clark 1984). Much work has indicated that negative affectivity is a potential confounding factor in associations between stress and illness reports (Costa and McCrae 1985, 1987, Watson and Pennebaker 1989, Ellington and Wiebe 1999). These associations have been interpreted as resulting from a tendency to scan the environment for threatening stimuli and interpret ambiguous stimuli in a negative manner (Watson and Pennebaker 1989). Cameron et al. (1998) also found that trait anxiety in breast cancer patients in remission was associated with increased levels of cancer worry. It is possible therefore that negative affectivity may be associated with measures of both general and cancer distress as well as representations of breast cancer and act as a confounding factor in this research. In the most extreme case it is possible that high levels of distress in women at increased risk of breast cancer reflects a general dispositional style to perceive situations in a negative manner and this in turn may lead to them to develop negative representations of the disease. Additional research utilising adequate controls or longitudinal designs are required to explore this issue in more depth. The confounding impact of negative affectivity in this research will be discussed in more detail in Chapter 11 (see 11.3.2, page 320).

Baron and Kenny (1986) also report that using multiple regression to test mediation models assumes no measurement error of the mediator. Although the IPQ-R showed adequate psychometric properties in the increased risk sample the measures were not perfect with some subscales showing low internal consistency (see chapter 6). This will reduce the effect of the mediator and overestimate the effect of the independent variable on the dependent variable. An additional criticism of this analysis concerns the measurement of experience. Many of the experience variables were single items designed pragmatically to assess specific aspects of experience. Although these measures were useful indicators in this research it is possible that single items were not adequate to capture sufficient details of the experience for use in mediation analysis.

10.5.4 Conclusions

Overall the results showed that cognitive representations of breast cancer appeared to mediate the impact of recency of bereavement from breast cancer in the family on levels of general distress although this effect was not significant. In addition, emotional representations of breast cancer partially mediated the impact of aspects of subjective experience of breast cancer in the family on levels of cancer worry. Although the results must be interpreted with care due to the possible effect of negative affectivity, measurement error and conceptual overlap of constructs, the results are encouraging. Cognitive and emotional representations of breast cancer may be important mediators to target in interventions aimed at reducing levels of general and cancer specific distress respectively. These results support further work to apply this model to understanding variations in distress in women at increased risk of breast cancer and the development of interventions.

CHAPTER 11
DISCUSSION

DISCUSSION

11.1 SUMMARY OF MAIN FINDINGS

The research reported in this thesis was designed to improve understanding of the variation in levels of distress among women with a family history of breast cancer. It was proposed that personal experiences of breast cancer in the family were related to distress and that this effect was mediated by illness perceptions. The main findings in regards to the aims and predictions outlined in Chapter 4 are summarised in Table 11.1.

Table 11.1- Summary of main findings

Step in mediation model	Hypotheses	Findings
Step 1. Experience and distress (see Chapter 7. Summary and Discussion, Section 7.5, page 218)	• Women with a significant family history of breast cancer will show higher levels of distress than women without any experience of breast cancer.	Women at increased risk reported higher levels of cancer specific distress than controls. There was no difference in levels of general distress between the samples.
	• Experiences of breast cancer in the family will be associated with levels of distress in women at increased risk.	Both general distress and cancer specific distress were associated with experiences of breast cancer in the family. General distress was associated with issues relating to bereavement and cancer specific distress was associated with subjective elements of the experience.
	• Experiences of breast cancer reported by women in the general population will be associated with levels of distress in this sample.	Experience of breast cancer in the general population sample was associated with cancer specific distress but not general distress. Recent experience of breast cancer in friends or family was positively associated with cancer specific distress.
Step 2. Experience and illness perceptions (see Chapter 8. Summary and Discussion, Section 8.5, page 250)	• Women with a significant family history of breast cancer will hold more negative perceptions of the disease than women without any experience of breast cancer.	A larger proportion of women at increased risk held negative representations of breast cancer compared to controls. Women at increased risk had a more coherent understanding of breast cancer; a stronger emotional representation of the disease, and were more likely to believe the disease holds greater consequences than controls.
	• Experiences of breast cancer in the family will be associated with perceptions of the disease in women at increased risk.	The overall pattern of representations (as indicated by the illness representation clusters) and individual dimensions of illness representations were associated with specific experiences of breast cancer in the family.

	• Experiences of breast cancer reported by women in the general population sample will be associated with perceptions of the disease.	The overall pattern of representations was not associated with experience of the disease in the general populations sample. Any experience of breast cancer in the general population was associated with a more coherent understanding of the disease, perception of breast cancer as holding greater consequences. A recent experience of breast cancer in friends or family was associated specifically with stronger emotional representations and a perception of the disease as long lasting.
Step 3. Illness perceptions and distress (see Chapter 9. Summary and Discussion, Section 9.5, page 277)	• Perceptions of breast cancer will be significantly associated with levels of distress in women with a significant family history of breast cancer.	Levels of general distress and cancer worry in the increased risk sample were associated with a number of dimensions of illness representations (identity, timeline acute/chronic, consequences, emotional representations). Cancer worry was also associated with the overall pattern of representations (as demonstrated by the illness representation clusters). Intrusive thoughts about breast cancer were associated with emotional representations and perceptions of treatment control and timeline acute/chronic. In the control sample fewer and weaker associations between illness representations and distress were identified.
Step 4. The mediation model (see Chapter 10. Summary and Discussion, Section 10.5, page 302)	• The impact of experience of breast cancer on levels of distress will be mediated by perceptions of breast cancer.	Differences in cancer worry between the increased risk and control sample were found to be partially mediated by scores on the emotional representations and consequences subscales of the IPQ-R. In the increased risk sample the impact of recency of bereavement on general distress was found to be mediated by scores on the identity and consequences subscales of the IPQ-R. The emotional representation subscale was found to mediate associations between a number of subjective experience items on cancer worry.

The first step in testing the mediation model was to explore the relations between experiences of breast cancer and level of distress (Chapter 7). This work built on pilot work that had identified associations between quantitative measures of experience and measures of distress (Chapter 6). The results confirmed the hypothesis that women who had different levels of exposure to breast cancer in their social environment showed different levels of distress. Women with a significant family history of breast cancer and women in the general population who reported a recent experience of breast cancer showed higher levels of cancer specific distress than women with no experience of the disease. Particular experiences of breast cancer reported by women at increased risk were associated with levels of both general and cancer specific distress. Case level distress was associated with recency of bereavement of a family member from breast cancer and the number of personally known relatives to been affected with the disease. Cancer specific distress was associated with a number of experience items that reflected the subjective impact of the experience (eg its effect on family roles and life plans). Experience items were found to be significant predictors of both general and cancer specific distress. However, the proportion of variance accounted for was not large suggesting that there were likely to be other factors to consider.

These results contribute to the debate concerning level of general distress in women at increased risk of breast cancer. Previous research has been contradictory with some studies reporting higher distress in women at risk of breast cancer (Kash et al.. 1992, Gagnon et al. 1996) and others reporting levels comparable to normative scores (Wellisch et al. 1991, Lerman et al. 1994a, Lloyd et al. 1996, Zakowski et al. 1997, Coyne et al. 2000) (See Chapter 1, 1.4, page 48). This study found that a significant family history of breast cancer is sufficient to increase anxiety and concern specifically related to the disease but is not always associated with increased general distress. It is possible that previous research concerning levels of distress in women at increased risk research may have been inconsistent because the samples differed on factors (eg bereavement) related to general distress. Experiences of breast cancer in the general population sample were also related to cancer specific distress. This provides additional support for the notion that women's experiences of breast cancer are important predictors of distress regardless of risk status.

The second step in the mediation model was to examine associations between experience of breast cancer and illness perceptions (Chapter 8). In line with predictions based on the SRM women with different levels of exposure to breast cancer were found to hold different cognitive and emotional representations of the disease. Women with a family history of breast cancer were more likely to hold an overall negative representation of the disease compared to women without any breast cancer experiences. In addition, specific experiences of breast cancer were associated with illness perceptions in both women at increased risk of breast cancer and women in the general population. This suggests that a significant family history of breast cancer is associated with more negative perceptions of the disease but that certain experiences of breast cancer are associated with specific illness representations. Both negative and positive experiences were found to be associated with illness perceptions. This confirms previous qualitative studies that identified links between experiences of cancer and perceptions of the disease (Dudok deWit et al. 1997, Lodder et al. 1999, Leedham and Meyerowitz 1999).

The third step in the mediation model was to test for associations between illness perceptions and levels of distress (Chapter 9). Women at increased risk of breast cancer who held overall negative representations of the disease showed higher levels of general and cancer specific distress. A number of dimensions of illness perceptions were associated with levels of distress in women at increased risk of breast cancer in the predicted manner. Fewer and weaker associations between illness perceptions and distress were identified in the control sample suggesting that the link between illness perceptions and distress in healthy individuals is more salient for those at risk of developing the disease. Multiple regression analysis confirmed that illness perceptions predicted level of distress in women at increased risk of breast cancer in combination with experience variables although the amount of variance accounted for was still fairly small.

These analyses supported the premise of mediation. The final stage of analysis involved identifying and testing potential mediation models using multiple regression analysis (Chapter 10). Although a number of potential mediation models linking

experience, illness perceptions and distress were identified in the increased risk sample few were supported by the analysis. The results suggested that higher levels of cancer worry in women at increased risk of breast cancer compared to women with no experience of the disease was found to be partially explained by women at increased risk holding stronger emotional representations and perceiving the disease as holding greater consequences. In the increased risk sample, recency of bereavement was associated with stronger perceptions of the identity and consequences of breast cancer that accounted for higher levels of general distress. Previous research had identified links between parental death from cancer and psychological wellbeing in women at increased risk of breast cancer and suggested these relations may be mediated by perception of risk although tests of this model have been inconclusive (Zakowski et al. 1997, Erblich et al. 2000). This study suggests that perceptions *of the disease* may be the crucial factor.

11.2 THEORETICAL IMPLICATIONS

The work in this thesis was guided by the Self Regulatory model. Since this model was developed primarily to understand patients response to illness constructs reflecting perceived risk are not explicitly incorporated in the model. Other models in health psychology such as the Health Belief Model (HBM) (Rosenstock 1996, Becker 1974) have been designed in order to understand preventative health related behaviours and include concepts of perceived vulnerability and susceptibility (see Chapter 3, section 3.3, page 75). These constructs are likely to be important in understanding and explaining response of individuals to disease risk. However the HBM was designed to explain decision making and behaviour rather than emotional response that was the focus of the current research. Indeed the inability of the HBM to incorporate of emotional factors is a major critique of the model. The model is static in nature and does not identify how cognitions are formed or changed. In addition apart from providing cues to action the model does not account for the impact of social factors on cognitive representations.

The SRM was chose because of its focus on both cognitive and emotional representations and the ability to understand a range of psychological outcomes including psychological wellbeing. The model was felt to examine in more detail the

meaning of risk for the individual and explicitly identified the potential link between experience (stimuli) and development of illness perceptions. This theoretical grounding as well as the qualitative and anecdotal evidence linking experience of cancer and illness perceptions (Chapter 2) provided a strong justification for the current research. Future research might usefully consider the additional constructs of perceived susceptibility and potential interactions between this construct and illness representations in determining psychological response to risk.

These studies were amongst the first to have applied the SRM to understanding psychological response in individuals at increased risk of disease. The results support the premise of the SRM - that illness perceptions are derived not only from direct somatic or symptomatic experience but also from information available in the external social environment (Leventhal et al. 1980, Leventhal et al. 1984). Being at increased risk of breast cancer is sufficient stimulus to evoke representations of the disease and different experiences of breast cancer in the family are associated with different illness perceptions. Applying this model to women at increased risk of breast cancer can enhance our understanding of why some women report more distress than others for the same level of genetic risk. Experiences of breast cancer and illness representations were both found to be associated with and predictive of levels of general and cancer specific distress. The results therefore inform us of not only of factors that contribute to distress in women at increased risk of breast cancer but also how these factor interrelate.

The associations between illness perceptions and psychological well-being in the increased risk sample mirror to some extent findings from samples of symptomatic patients. Studies from a range of patient groups have shown beliefs concerning consequences; identity and timeline are negatively associated with psychological adaptation to illness (eg Heijmans and de Ridder 1998, Scharloo et al. 1998, Hagger and Orbell 2001). The same associations were demonstrated in women at increased risk of breast cancer but not women in the control group suggesting that a sense of threat is a prerequisite for this effect. Reliable associations between the identity dimension and psychological well-being were however found in both samples. This confirms that the identity dimension is not merely tapping the symptom *experiences*

of patients but rather *beliefs* concerning nature of the illness in question (Moss-Morris et al. 2002). These findings also support the notion that identity is an important component of illness representations for healthy individuals (Bishop et al. 1987). Bishop et al. (1987) proposed that lay conceptions of illness are based primarily around symptoms in order to aid detection of disease. The results from this work suggest that additional representations are also important in healthy individuals *at risk* of disease and may reflect the personal meaning of this risk.

Not all predictions derived from the SRM were met in this research. Few associations between control beliefs and psychological well-being were identified. This was surprising since associations between control beliefs and adaptation to illness have been well demonstrated in patient populations including breast cancer patients (eg Heijmans and de Ridder 1998, Scharloo et al. 1998, Hagger and Orbell 2001, Taylor et al. 1984, Buick 1997). It is likely that beliefs concerning personal control *over risk*, such as ability to prevent breast cancer and confidence in screening methods are more important in this population than control *over the disease* as assessed by the IPQ-R. Access to information about breast cancer prevention has been identified as an important information need for women at increased risk of breast cancer and one of the main reasons why women attend familial breast cancer clinics (Appleton et al. 2000, Brain et al. 2000). This suggests that women are actively developing representations regarding personal control over their risk. Chalmers and Thompson (1996) found the process of adapting to risk involved developing a sense of personal control. Lerman et al. (1996) also suggested that genetic counselling promotes psychological adjustment by improving knowledge about breast cancer and potential for prevention/early detection.

However, few studies have looked at beliefs regarding control of risk or prevention of breast cancer in individuals at risk. A study by Stewart et al. (2001) indicated that beliefs regarding personal control over breast cancer recurrence are widely held by survivors of the disease (see section 3.4.6, page 90). Audrain et al. (1997) assessed beliefs about control over developing breast and ovarian cancer in 256 women who had self-selected for genetic counselling. Based on one single item used to assess beliefs about control over developing breast cancer, 55% of women reported no or

little control and 45% perceived at least moderate amount of control. Women who were classified as having higher perceived control over developing breast cancer were found to show lower levels of general and cancer specific distress. An interaction indicated that general distress was higher for women with low perception of control and high perception of personal risk. Although this suggests that beliefs about control of risk are important, it is difficult to elucidate the meaning and interpretation of one single generic item given the range of issues regarding control in this population.

The SRM needs to be expanded to encompass beliefs regarding control over risk when applied to individuals at risk of disease such as breast cancer. Control over breast cancer in healthy individuals is a complex issue encompassing a number of elements. There is a need to consider beliefs about prevention and risk reduction. Perceptions of personal control (eg behaviour) as well as beliefs regarding the efficacy of screening and benefits of early detection need to be explored. As well as beliefs regarding control of developing breast cancer it may also be important to consider women's beliefs about control over disease progression and risk of dying from breast cancer if they were to develop the disease. Horne (1997) provided convincing evidence that beliefs about treatment and medicine were an important component of lay representations. No study has directly investigated these beliefs although qualitative studies have indicated that healthy women hold perceptions of breast cancer treatment (Payne 1990, Savage and Clarke 1998, see section 3.4.7, page 90). Women's perceptions of specific treatments for cancer as well as beliefs about the potential advances in treatment may be important aspects to consider. Further exploration of these constructs and investigation into how they may fit within the SRM would be a worthwhile contribution to research in this area.

The SRM is dynamic and suggests that illness representations and coping mechanisms related to risk are not static elements but change depending on stimuli and cues in the environment. Applying the SRM to women at increased risk of breast cancer may not only help us to understand individual differences in psychological response to risk but also variation in psychological well-being over time. Easterling and Leventhal (1989) suggest that individuals at increased risk of a life threatening

disease are unlikely to worry continuously but reminders of the risk will prompt concern and anxiety. Events in the family (eg diagnosis, relapse or bereavement) are likely to invoke distress and activate as well as alter representations. Clinic appointments and the prospect of screening might also activate illness representations and induce distress. Easterling and Leventhal (1989) found ex-patients worry about breast cancer was reactivated during visits to their doctor and qualitative studies of women at increased risk of breast cancer identified heightened distress and anxiety at the time of clinic appointment (Appleton et al. 2000).

> *"I don't think of having breast cancer until it becomes like the end of May, beginning of June when I know that my appointment is going to pop through the letter box that's actually when I actually start to think more about it and it becomes into the forefront in your mind if you like".* (Appleton 1999, quote from telephone focus group study, personal communication).

Further research in this area may hold clinical application as well as enhance understanding of the dynamic interactions within the SRM.

The results reported in this thesis have shown how the SRM can act as a useful framework to enhance understanding of how factors contribute and interact to influence psychological response to breast cancer risk. A recent study using alternative methodology has reported findings that are supportive of the mediation model. McAllister (2002) presented a grounded theory based on interview data to explain response to genetic testing for Hereditary NonPolyposis Colorectal Cancer (HNPCC). She found that individuals' experiences of cancer in their family influence the degree to which they 'engage' with their risk. Engagement was a concept derived from the data that reflected '*the degree of cognitive and emotional involvement with one's increased risk of developing cancer as a result of one's family history of cancer'* (McAllister 2002, page 8). Level of engagement is then proposed to influence response to testing. Individuals who were 'intensely engaged' tended to have witnessed closer family members suffering or dying from cancer. This theory was developed from qualitative analysis of interviews without theoretical guidance. However there are parallels between the concept of engagement and activation of illness representations in individuals at increased risk of cancer because of their

family history. Greater understanding of the overlap between these concepts and models may assist further theoretical development in this area.

11.3 METHODOLOGICAL ISSUES

11.3.1 Sample

It was important that the samples obtained for this work were representative in order to be able to generalise the results and conclusions of this study. A random selection of women at increased risk of breast cancer was therefore selected from the database held at the clinical genetics department. It was also important that the general population sample was comparable to the increased risk sample on factors that may influence knowledge and perceptions of breast cancer (eg educational level). In order to attempt to achieve this the general population sample was selected on the basis of information concerning age and postal region of the increased risk sample. Within the general population sample a quarter of individuals reported that a relative had experienced breast cancer. It is possible therefore that a small proportion of these individuals may have an increased (genetic) risk of breast cancer themselves. This justified use of a control sample who had no experience of breast cancer in their social environment as the main comparison group.

It was only possible to recruit one sample for this study due to ethical constraints on research in the increased risk sample (see section 54.6, page 110). Subsequent methodological problems also arose in accessing general population samples. Following recruitment for this study, Lothian Health Board implemented new confidentiality guidelines that restricted researchers from accessing the Community Health Index to obtain details on individuals within the region. Although the study was carefully designed to test a number of hypotheses the reliability of the results are unknown and it is not possible to generalise findings from a single sample.

The samples were likely to have been biased to some extent. The samples were predominately white, Caucasian and of a high educational level as seen in many studies of screening populations (Rimer et al. 1996). Women who seek genetic testing have been found to be of higher education and socio-economic status (Codori

et al. 1994, Kash et al. 1997). The comparison of respondents and non-respondents reported in section 5.3.2 (page 120) also revealed a trend for respondents to come from areas of lower social deprivation. Individuals with higher levels of education are more likely to hold beliefs about breast that are compatible with scientific and medical approaches (Bowling 1989). It is possible that the general population sample was also biased towards those interested in breast cancer and health issues. It is unlikely therefore that the results of this study can generalise to women of lower educational level or ethnic groups whose beliefs about breast cancer may be diverse (Klonoff and Landrine 1994).

The sample may also have been biased in terms of levels of distress. Women with high levels of distress may avoid cues about their risk, including participating in research studies. Indeed scores on the general distress and Impact of Event Scales were positively skewed in the increased risk sample. The lack of associations between avoidance as measured by the Impact of Event scale and experience may reflect that women avoiding breast cancer issues did not complete the questionnaire. This suggests that women with the highest levels of distress whom we are most keen to understand may not have been captured with this research. Although avoidance may be maladaptive there are also cases when avoidance has been shown to be a successful coping strategy for dealing with breast cancer (Cordova et al. 1995, Primo et al. 2000). Without access to the subgroup who did not participate in the research it is impossible to determine if this reflects higher distress or an adaptive coping strategy.

Women in the increased risk sample had previously made the decision to seek information about their risk and had obtained genetic risk counselling. This suggests that these women were actively coping with their risk. It is possible that women with a family history of breast cancer who choose not to attend for genetic counselling may hold different representations of the disease. In addition, the experience of attending for genetic counselling may have influenced both illness representations and levels of distress. Different results may therefore be obtained in women prior to genetic counselling. The experience of genetic counselling may also differ between centres or between genetic counsellors. Further research is therefore required to

investigate the reliability of effects described here in other familial cancer clinics and to examine illness representations at different time points.

Comparisons were made between women at increased risk of breast cancer and women in the general population in order to determine differences in distress and illness representations between samples with different levels of experience of breast cancer. However, the samples also differ on experience of genetic counselling for breast cancer. It is impossible to conclude from this study that differences between the samples are a consequence of experience of the disease in the family alone. Such differences may also be attributed to information obtained in genetic counselling and awareness of risk status. A controlled prospective study examining illness perceptions before and after genetic counselling would be required to investigate this issue.

11.3.2 Design

The study was designed using the SRM as a theoretical framework in order to examine associations between experience, illness representations and psychological wellbeing in an 'at risk' sample. Although the results from this work supported the application of the SRM to this new population, the study was cross-sectional and conclusions regarding causality can not be made. A number of alternative plausible interpretations can be provided. One of the major limitations in this study and health psychology research in general is the confounding influence of negative affectivity. It is possible that negative affect influences levels of distress, reports of past experience and illness perceptions and creates a conceptual overlap between the measures used within this study. In this way negative affect may account for the relationships reported throughout this thesis.

Negative affect is conceptualised as either a state or trait. Trait negative affect refers to a stable underlying personality factor that is associated with negative mood and self concept (Watson and Clark 1984). State negative affect reflects temporary fluctuations in negative mood. Trait negative affect has been shown to influence the interpretation of stimuli (Watson and Clark 1984, Watson and Pennebaker 1989) and

may therefore confound the measures utilised in this research. Surprisingly little work has addressed the impact of negative affectivity on illness representations with the exception of Taylor et al. (1991) who provided preliminary evidence to suggest that association between feelings of control over disease and adjustment to illness was not confounded by negative affectivity.

Studies of depressed patients have shown negative retrospective biases in clinical populations (Beck et al. 1979). Experimental research from cognitive psychology has shown that transient induced mood states result in recall patterns similar to those shown by depressed patients and increased anxiety has been associated with greater retrieval of threat related events from autobiographical memory (Sutton et al. 1988, Healy and Williams 1999, MacLeod 1999). These findings have led to a number of theories regarding the impact of mood states on cognitive processes including memory (reviewed by Ellis and Moore 1999). Based on clinical observation, Beck (1979) proposed the schema theory that suggests current mood states guide the processing and organisation of information. Sad or depressed individuals are thought to be subject to a maladaptive 'depressive schema'. This consists of sets of beliefs and assumptions that produce negatively distorted thinking and assists retrieval of mood related memories (Beck 1979). Following experimental work investigating cognitive processes and affective states, Bower (1981) outlined the 'associative network theory' in which mood states are proposed to be represented semantically in memory. Affective states increase the accessibility of negative memories, concepts and representations resulting in mood dependent thinking. Although these theories have been extended and more comprehensive theories proposed, the link between emotional state and cognition is clear (Teasdale 1993, Ellis and Moore 1999). It is possible therefore that negative affective states in women at increased risk of breast cancer might not only assist retrieval of negative events from memory but also evoke more negative interpretations of the experience and representations of breast cancer. In this way negative affectivity may influence all self report data in this study and account for the observed relationships. Future research needs to examine associations between personality dispositions and illness representations and attempt to control for factors such as negative affectivity.

Causality between cognition and emotion has been debated for many years (discussed by Lazarus 1999). A main advantage of using the SRM in this area is that it raises awareness of *both* cognitive and emotional representations of breast cancer and potential interactions between these levels of processing. However, the multivariate and transactional nature of the SRM makes it a difficult model to test. The researcher must conceptualise and measure multiple potentially overlapping factors as well as specify the independent and dependent factors at a certain points in time (Leventhal and Cameron 1987). The dynamic appraisal processes in the model suggests that all factors are to some extent interrelated in a discursive manner. Although this makes causality difficult to examine it is realistic to accept continuous reciprocal causality between cognitive and emotional constructs (Lazarus 1999). The research reported in this thesis has therefore been worthwhile to indicate the cognitive representations involved in emotional response to breast cancer risk even if causality has not been established. In depth qualitative studies, as well as intervention studies and prospective longitudinal studies assessing the impact of illness related events on illness representations and psychological well-being will provide further insight into the dynamic processes involved. These designs may provide further insight into the causal relations between experience, illness perceptions and psychological response to risk. For example a longitudinal study may be able to determine the impact of genetic counselling on illness perceptions and distress. Such a design will be able to determine what illness perceptions are held prior to genetic counselling and how they are associated with distress; the impact of genetic counselling on illness perceptions and level of distress; what changes in illness perceptions are associated with psychological wellbeing in the longer term.

11.3.3 Measures

11.3.3a Experience

The results reported in this thesis suggest that it is possible although difficult to measure experience of breast cancer in the family in a quantitative manner. The problems appeared to arise from attempting to encapsulate a vast range of experiences important in this context in a short questionnaire. The concept of experience of breast cancer in the family is wide and the questionnaire aimed to

capture important aspects of women's experiences that might influence their response to their own risk. However, the face validity of the questionnaire is dubious. In particular some items (eg resemblance items) do not refer directly to experiences of breast cancer but rather perceptions of similarity to the affected relative.

Although focussing on the index relative appeared to be a useful and workable format a number of problems arose during analysis of these items. A few respondents reported that they were too young to remember any of the experience surrounding their index relative and omitted the questions. Some items were also found to hold multiple answers. For example, one participant reported that although her mother was first diagnosed when she was 6 years old her breast cancer recurred when the participant was age 27. Although the experience items asked for participants to report when breast cancer was *first* diagnosed in the index relative it is likely (although unclear) that respondent referred to the more recent experiences in subsequent questions. It is likely that women in families with more than one affected relative have witnessed a range of different outcomes and consequences of breast cancer and did not have the opportunity to express different or conflicting experiences in the questionnaire provided. The quote below highlights these dilemmas. In addition, women with a family history of breast cancer may be more sensitive to breast cancer experiences in individuals outside the family but this is not captured in the questionnaire.

> *"My mother has had a second bout of breast cancer and this time it had spread to her spine and this has brought it home more, because I am seeing her slowly losing her mobility and its inoperable and it will be terminal, and you know, although the first time she had breast cancer it all seemed, you know treated very effectively and we thought that she was cured. This second occurrence and the after effects of it have made me more concerned, I think with the realism"* (Appleton 1999, quote from telephone focus group study, personal communication).

Despite a number of qualitative studies reviewed in Chapter 2 no quantitative measure of experience was available in the literature. The measure used in this study was under development within the research group and based on interviews and clinical observations with women attending the familial breast cancer clinic. The pilot analysis showed the measure to be acceptable to women at increased risk of

breast cancer and also revealed predicted associations with levels of general and cancer specific distress. Since the aim of this research was to test the mediation model (Fig 4.1, page 100) there was insufficient time to develop and validate a measure of experience. The current questionnaire was therefore deemed the most appropriate available instrument to use during this study. However, the face validity and reliability of the questionnaire remains a strong limitation of this research. In particular, the use of single items to assess particular experiences may have resulted in unreliable measures. Although items were reduced using scaling analysis this analysis was pragmatically rather than conceptually driven. Studies assessing experience in a different manner may therefore produce different results. Future research in this area may proceed by focussing on specific aspects of the experience (ie exposure to breast cancer in the family, changes to life created by breast cancer) rather than attempting to assess a broad range of experiences. In this way measures of specific experiences can be developed. Alternatively, a checklist of experiences that contribute to heightened distress could be developed. This could comprise of a list of categorical questions (eg have you experienced a bereavement from breast cancer in your family?) that could be summed to provide a score that reflects the degree of breast cancer experience within a woman's family.

Although the questionnaire was designed to assess aspects of experience that had been highlighted as important in this population and were hypothesised to be mediated by illness perceptions, a number of pertinent aspects of experience were neglected. These included details of the illness experience (how breast cancer was detected, treatment regime, disease progression and recurrence) involvement with care-giving, stage of the family and concurrent stressors (Northouse 1995, Siegel et al. 1996, Veach and Nicholas 1998, Lewis et al. 1993).

The experience questionnaire focused on past experiences in the family. It may be difficult to identify associations between past experience and measures of current anxiety such as intrusive thoughts or avoidance. Future research could be directed at understanding the effect of current breast cancer experiences. It may be useful to compare women who currently have a relative in their family who is suffering from breast cancer to a matched control sample of women at increased risk who are not

experiencing any breast cancer episodes in their family at the time. This would help reduce the impact of negative retrospective bias and allow more insight into the experience and its effects. Prospective longitudinal studies also would provide clearer insights into how experiences impact on emotional response to risk and are necessary to examine causality. However, great care would need to be taken when conducting research at such a sensitive and difficult times.

11.3.3b Distress

This study and the majority of research assessing distress in women at increased risk of breast cancer used self-report screening measures. These measures are designed to be inclusive and therefore overestimate the rate of actual psychiatric morbidity (Hopwood et al. 1998, Coyne et al. 2000). It has been noted that there is often a lack of distinction between distress and clinical disorder throughout the literature in this area (Coyne et al. 2000). This study was designed to understand factors that contribute to general distress rather than psychiatric disorder and therefore the GHQ was deemed adequate. In addition a control sample enabled direct comparison with an appropriate population. Studies that wish to look specifically at rates of clinical disorder or to assess psychological support needs would be best advised to follow up screening measures with interviews and ensure comparisons with controls or relevant normative data.

The results from this thesis was consistent with research in the literature indicating the importance of cancer specific distress in this population (eg Lloyd et al. 1996). The use of cancer specific distress measures is now widely accepted in order to provide information about the nature of distress and inform interventions (Thewes et al. 2001). However, there are concerns that these measures may reflect a realistic response to the situation rather than morbid anxiety (Coyne et al. 2000, Hopwood et al. 2001). Cancer specific distress and concern about breast cancer was also found to be prevalent in the general population of women of this age. This is the first study to have reported levels of cancer worry in the general population. It is possible that the sample in this study were biased to women with experience of breast cancer. Sixty-one percent of women reported to have some experience of breast cancer in their social environment and 25% reported that a relative had suffered from breast cancer.

It is possible that women with experience of breast cancer were more likely to respond to the questionnaire and show higher level of cancer worry than women without such experience. However there was no difference in the level of cancer worry reported by women in the general population sample with and without experience of breast cancer in their social environment. Further research is needed to further examine the level of cancer worry in general population samples. It is not unexpected that women in the general population would show cancer worry given the high profile of breast cancer and promotion of self care practises (BSE) in the media as well as the prevalence of health related intrusive thoughts in healthy samples (Freeston et al. 1994). Although women in the increased risk sample showed higher cancer worry than controls it is worth noting that nearly 50% of the increased risk sample reported not to have thought about breast cancer in the previous week. This perhaps raises concerns about the meaning and clinical utility of the cancer worry scale. Further work is needed to examine the clinical significance of this measure and to provide normative data.

Using the Impact of Event scale in samples with differing levels of distress was problematic. Although the opt out box was useful to exclude women who had not thought about breast cancer in the past week, it reduced the sample size below that required for the analysis plan. It also made the results difficult to compare with other published studies which did not include the opt out box in the questionnaire. Although the Impact of Event scale was designed to allow the sub-scales to be summed the intrusion and avoidance subscales were analysed separately in order to understand the contribution of experience to these constructs. This was justified by the different patterns of associations identified. Primo et al. (2000) argues that the association between these constructs and adjustment to breast cancer may be even more complex and that patterns of intrusion and avoidance over time may prove to be better indicators of adjustment. This would require a longitudinal investigation. Whilst the Impact of Event scale was useful to assess the magnitude of intrusive thoughts it does not provide any information regarding the content of these intrusions. It would be clinically important to investigate the nature of intrusive thoughts in women at increased risk of breast cancer and whether specific thoughts or images provoke greater anxiety.

11.3.3c Illness perceptions

Two major aims of this thesis were to extend the SRM to individuals living with an increased risk of breast cancer and to test the predictions of the model in this sample. In order to achieve this it was necessary to adapt a generic quantitative measure of illness perceptions that had been developed and used widely in patient populations – the IPQ-R (Weinman et al. 1996, Moss-Morris et al. 2002).

Not all components of illness representations are likely to be relevant to all diseases and it was expected that not all dimensions assessed by the IPQ-R would be appropriate components of healthy women's beliefs about breast cancer. In addition it is likely that other psychological constructs such as perceived susceptibility and vulnerability from other theoretical models such as the Health Belief Model (are likely to be important cognitions to consider (Rosenstock 1996, Becker 1974).

One subscale in particular (timeline cyclical) was found to be inappropriate for use in this population and omitted from subsequent analysis (see 6.19.1, pages 171-174). Whilst some subscales (eg emotional representations and illness coherence) showed high internal consistency in both samples other subscales were found to show different levels of reliability in women at increased risk and women in the general population suggesting that certain dimensions of illness representations may be more or less relevant depending on women's risk status or experience of the disease. In the general population sample, the identity and consequences subscales showed the highest levels of internal consistency of the original 5 cognitive dimensions. The timeline acute/chronic and control subscales showed lower internal reliability. This is consistent with Bishops work on disease prototypes in healthy individuals. Bishop et al. (1987) found that healthy individual's comments about serious diseases most often reflected representations of the label, cause and consequence of disease and less often cure or timeline. Although the quantitative measures derived from the IPQ-R is able to inform us about the strength of illness perceptions in these samples it does not give us any information regarding the relative importance of dimensions or how elaborated the illness perceptions are in each sample. Further qualitative analysis would be required to explore these issues.

There were initial concerns that the identity scale would be difficult to apply to breast cancer because of the limited symptoms associated with the disease (see Chapter 1). Also, since women were not referring to their own illness the scale could be interpreted as reflecting on a spectrum of symptoms ranging from early breast lumps to symptoms related to advanced metastatic disease. It is possible that some women may have reported symptoms of breast cancer prior to diagnosis whereas others may have included treatment side effects when responding to this scale. This may have increased the number of symptoms reported on the identity subscale and the variability of scores obtained. In the future it may be beneficial to explicitly state if the scale is referring to symptoms alone or symptoms resulting from both the illness and its treatment.

A main issue that became apparent during analysis of the results was the conceptual overlap between cancer specific distress measures and emotional representations of breast cancer as assessed by the IPQ-R. The high correlation between these constructs provided information regarding the concurrent validity for this new subscale however, such strong associations may lead one to argue that the emotional representations subscale of the IPQ-R could be construed as a measure of cancer specific distress. Although high correlations would be expected between these measures, cancer specific distress represents more than affective response to breast cancer alone. It also reflects persistent thoughts about developing the disease and the impact of these concerns on daily functioning.

Although the IPQ-R proved adequate for use in this thesis there is room for improvement and further work needs to explore the validity of the scale in women at increased risk of breast cancer. In retrospect, it would have been informative to have examined the face validity of the questionnaire in more depth at the piloting phase. The questionnaire was piloted on women at the clinic. It is possible that these women were concerned about their clinic appointment and anxious about screening (Wardle and Pope 1992, Appleton et al. 2000) and were therefore not in an ideal position to concentrate on reading and comprehending a questionnaire. It would have been more systematic to have piloted the questionnaire in a postal format to a random sample of women at increased risk of breast cancer and to have included an evaluation sheet for

women to complete. Alternatively, a qualitative assessment of women's understanding of the purpose, relevance, acceptability and comprehension of the measure would have provided useful data on the face validity of the IPQ-R in this population. This approach has been used previously when adapting questionnaires for this population (Thewes et al. 2001).

Qualitative methodology could have been used to further investigate the concurrent validity of the measure. It would have been informative to have conducted interviews with a proportion of the increased risk sample and elicit illness representations in the manner outlined by Leventhal and Nerenz (1985). Data derived from these interviews could then be compared with responses on the IPQ-R and similarities and differences between the methodologies examined. This triangulation could be used to highlight items or dimensions of the IPQ-R that are less relevant to this population. This approach could also be used to explore the validity of patterns of illness representations identified by the cluster analysis and generate further information on the importance of particular representations in this population.

11.4 STATISTICAL METHODS

The methodology used in this research was driven by the need to test a mediation model. Quantitative measures were chosen in order to utilise the technique outlined by Baron and Kenny (1986). Few significant meditation effects were identified. On reflection the mediation model outlined in Chapter (Figure 4.1, page 100) may have been too broad. In the future it may be helpful to focus the model to test specific hypotheses concerning the mediation of specific experiences on particular psychological outcomes. The lack of mediation effects may reflect limitations of this technique or problems with measures (see Chapter 10, section 10.5.3, page 304). Poor measurement of the mediator leads to an exaggerated relationship between the independent and dependent variables. In this study a number of newly developed/adapted measures with limited psychometric testing were used. This may underestimate the mediation effects. Further techniques such as structural equation modelling that can be used in an attempt to explore associations between a range of variables (Baron and Kenny 1986). In addition, structural equation modelling can be

used to estimate feedback in the model (a causal relation from the dependent variable to the mediator). This may provide further information regarding causal relations.

The majority of analyses reported in this thesis, including the mediation analysis were correlational in nature and conducted on cross sectional data. Although this analysis supported the model causality can not be inferred from this type of analysis. The mediation analysis can not directly test the causal ordering of the model but is able to determine if the data supports a plausible ordering. Experimental studies involving randomised manipulation of treatment are the only test for causality. However practical considerations render this difficult in this field. The independent and dependent variables in this case (experience and distress) can not be ethically or practically produced experimentally and therefore randomisation into experimental groups is not possible. Interventions aimed at changing illness perceptions may provide an experimental test of the model but to date these techniques have not been adequately developed or described.

Cluster analysis was successfully utilised to explore the patterns of illness perceptions and identified similar results to that reported in previous studies (Buick 1997, Heijmans 1999). The validity of the clusters was supported by further analysis that identified predicted associations between the clusters and both distress and experience measures in both samples. One of the benefits of cluster analysis is its applied clinical relevance (Hack and Degner 1999). Cluster analysis can aid health professionals to identify subgroups of individuals and make assumptions regarding their adjustment and how to intervene to enhance patient's wellbeing. However, cluster analysis is a procedure based on heuristics rather than statistical reasoning and there are no tests of significance to determine the number of groups in the data. It is possible therefore that the two clusters identified in these samples may be further divided into smaller clusters. Another limitation to the cluster analysis performed on this data was the omission of causal items from the analysis. It is possible that causal beliefs are an important component of illness representations that may contribute to response to risk in healthy populations. Further research is required to investigate causal models in women at risk of breast cancer in more depth and explore associations with psychological response to risk.

Throughout the analysis care was taken to avoid multiple testing. Hypotheses usually involved making comparisons between the increased risk sample and women in the general population without any experience of breast cancer. Although it would have been interesting to have compared the increased risk sample with the full general population sample this would have involved repeating analysis on a proportion of the respondents in this sample.

11.5 CLINICAL APPLICATION

11.5.1 Understanding and reducing distress

Women with a family history of breast cancer conveying comparable levels of risk exhibited varying levels of distress. Even after genetic counselling a proportion of women at increased risk of the disease had fairly high levels of distress and showed greater cancer specific distress than controls. This suggested that some women might benefit from psychological support in order to promote adjustment to risk status and that interventions are likely to more effective if targeted specifically at reducing cancer specific distress. Researchers in this field have previously suggested that genetic counsellors should focus on sources of distress such as past cancer related events in the family during genetic counselling (Watson et al. 1999, Hopwood et al. 1998). Watson et al. (1999) suggest that psychological support needs to be integrated into the genetic counselling service either by broadening the training of genetic counsellors or including mental health professionals within the clinical genetics team. However, no guidelines currently exist for how to achieve this. Given the tight schedule of the clinics to date the provision of psychological services needs to be carefully planned and targeted at individuals who are most in need. The results obtained in this study indicate that it may be worthwhile identifying those women who have had certain experiences associated with distress (e.g. having lost a relative to breast cancer recently) and to discuss adjustment difficulties or screen for psychological problems using a validated screening tool such as the GHQ. These women could be provided with relevant self-help material (eg how to cope with bereavement, how to cope with worry) and information about psychological services or referred for more detailed psychological assessment if deemed necessary.

Women at increased risk of breast cancer not only face trauma regarding past experiences in their family but often also multiple loss, as well as ongoing bereavement and concurrent trauma concerning their own risk. In a discussion about responses to multiple loss from AIDS, Nord (1996) highlighted that current theories about grief and trauma are inadequate for understanding issues of multiple and ongoing bereavement since *'Existing theories and interventions concerning trauma inevitably assume that the trauma occurred in the past creating some degree of distance from the event'* (Nord 1996, pg 404). Issues of grief and trauma therefore need to be addressed in a different manner in this population. It is possible that preparing women to cope with potential breast cancer related events in their family (including diagnosis of self or other family members, communication about breast cancer, changing family roles and bereavement) may help prevent psychological problems. Chalmers and Thompson (1996) identified that rehearsing how to cope with diagnosis helped women to cope with breast cancer risk. Research on terminally ill patients and their families has indicated that preparation for death is seen as extremely important for all involved (Steinhauser 2000, 2001). These techniques might also help prevent persistent grief described in this population (Hopwood et al. 1998).

The results reported in this thesis have shown that women's experiences of breast cancer in their family influence representations of breast cancer as well as having a direct impact on levels of general and cancer specific distress. This suggests that genetic counselling may benefit from giving attention to both the emotional aspects of the counselees experiences in their family as well as their beliefs about what breast cancer risk entails. It is possible that women may have developed beliefs about the prognosis and treatment of breast cancer based on misinterpretations of experiences of family members. Kelly (1987) provided an anecdotal report of a woman whose father had received surgery for bowel cancer the day after reporting abdominal cramps to his GP. The woman reported how she subsequently believed bowel cancer to be an extremely rapidly developing cancer and became distressed about her own risk. However, after learning that her father had actually suffered from abdominal discomfort and rectal bleeding for at least one year and that detection

techniques were available her anxiety was reduced. Beliefs based on past family experience may not be congruent with subsequent improvements in the detection and treatment for breast cancer. Moyer and Salovey (1996) outline changes in the treatment and meaning of breast cancer over the past 4 decades. They describe how treatment has evolved from radical mastectomy at the time of biopsy (prior to the 1970s) to a two stage surgical procedure today in which patients are more involved in their treatment, have time to consider a variety of less intrusive options and are offered the choice of breast reconstruction. In addition, society has become more open about cancer and a wide range of support is available to breast cancer patients today (support groups, self help literature etc). Given the impact of women's experiences of breast cancer on representations of the disease and potential for misinterpretation of experiences and outdated information it may be beneficial not only to provide women with information concerning cancer genetics and their risk status but also up to date information concerning the detection and treatment of breast cancer. In a recent telephone focus group study of women attending the familial breast cancer clinic in Edinburgh, women requested information on a variety of topics including breast cancer treatments and current trials in order to help them cope with their risk status (Appleton et al. 2000). This type of information may help those holding misperceptions of breast cancer to update and restructure their representations of the disease and reduce levels of anxiety. A psycho-educational intervention is currently being carried out for women at the Edinburgh familial breast cancer clinic and includes provision of information about the diagnosis and treatment of breast cancer (Appleton, personal communication).

Health professionals involved in genetic counselling need to be aware of the disparate beliefs of women attending the clinic and to recognise illness perceptions that may be indicative of distress. The cluster analysis reported in this thesis suggested that an overall negative perception of breast cancer might prove to be a useful indicator. These representations may interfere with the comprehension and interpretation of information provided during counselling sessions. It may be useful to train genetic counsellors to elicit and identify illness representations and to structure the information they provide in order that counselees can integrate it with their experiences and existing belief structure.

There is potential for interventions aimed at improving psychological well-being in women at increased risk of breast cancer to be based around modifiable factors in the SRM. However results from this thesis alone are not sufficient to determine what factors should be targeted in interventions or the most appropriate techniques to use. Further work is required to investigate these issues. There have been a number of intervention studies carried out with women with a family history of breast cancer although the aims of the studies and samples recruited have been diverse. Studies have included problem solving training to promote adherence to BSE and reduce distress in women with a recently diagnosed first degree relative (Audrain et al. 1999, Schwarz et al. 1998) and use of introductory videos concerning cancer genetics prior to genetic counselling on accuracy of risk perception, distress and understanding of cancer genetics (Cull et al. 1998).

A few studies are currently under development or in progress to provide psycho-educational material to women living with an increased risk of breast cancer in order to improve psychological adjustment to risk. Kash et al. (1999) report an ongoing randomised controlled trial of an intervention incorporating social support, education and problem solving in order to enhance knowledge, coping skills, adherence to screening and to reduce breast cancer anxiety. Preliminary reports of the 1 year follow up reveal that the interventions appears effective in reducing breast cancer anxiety and increasing knowledge about breast cancer (Kash et al. 1999). Wellisch et al. (1999) also report a pilot intervention including sessions focused on education regarding genetics, medical information concerning breast cancer and nutrition as well as a psychological session regarding experience of being at increased risk of breast cancer. The intervention was been found to reduce depression and anxiety but had no effect on resolution of grief. Esplen et al. (1998, 2000) developed an intervention also including both psychotherapeutic and psychoeducational techniques. The intervention was aimed at increasing women's ability to cope with their risk, screening and management decisions as well as to come to terms with their experiences in their family and to '*detoxify the threat of developing breast cancer*'. The intervention included an exploration of women's experiences of breast cancer in their family in order to reduce fears associated with the disease, dispel myths about

breast cancer and to clarify information concerning the disease. Women were also given the opportunity to talk to breast cancer survivors. A pilot study of this intervention indicated a reduction in distress, depression, anxiety and unresolved grief in those who participated.

These initial studies are promising and suggest that interventions may effectively promote adjustment in women at increased risk of breast cancer. However many of the studies have only been conducted in pilot form (Esplen 1998, 2000, Wellisch et al. 1999) and randomised control trials are necessary to fully test the effectiveness of the intervention. In addition, most of the interventions contain a wide range of techniques and hence the effectiveness of specific strategies is difficult to evaluate. The interventions conducted to date are also extremely intensive, conducted on small groups of individuals, over a long period of time at high cost. There is a growing need to be able to target these interventions to those with greatest need who are most likely to benefit or to identify effective strategies within the interventions to enable the development and provision of less intensive interventions.

Taking a theoretical approach to intervention design and evaluation will assist the identification of effective components within the intervention. The studies conducted to date have lacked theoretical input and hence the mechanisms by which the interventions produce effects remain elusive. The SRM is a useful framework to assist the development and evaluation of intervention in this area. Using this model allows the researchers to identify and target specific factors, hypothesise about the mechanism involved and develop informative evaluation measures. The intervention study reported by Esplen et al. (2000) may have successfully altered a number of processes outlined in the SRM that have been examined in this thesis. The intervention aided the participant to identify experiences of breast cancer in her family that were associated with distress, challenge cognitive and emotional representations of breast cancer, assisted appraisal of representations and coping strategies and introduced a number of coping strategies relevant to incidences or emotional reactions that may occur in the future. This is illustrated in quotes describing the intervention below. The effectiveness and evaluation of this intervention would be greatly improved by explicit hypotheses detailing effects the

intervention was designed to achieve and the inclusion of relevant evaluative measures.

> *" A goal of the supportive-expressive group therapy model is to help the women expand and improve their repertoire of coping skills. Discussions that focus on the problems encountered by group members and current coping strategies are promoted in the group...the group also teaches relaxation methods (such as guided imagery) to provide women with a behavioural technique that can facilitate their ability to cope with testing or screening procedures*" (Esplen et al. 1998, pg 378)

> *"Additionally, myths about breast cancer risk that may have evolved from a particular family experience or through the media can be dispelled, or information can be clarified. This is the key role of having a woman with breast cancer in the groups. There can be direct exchange between those at risk and those who have breast cancer. Women at risk can ask questions about breast cancer and treatment to those who are dealing with the illness. Women who had little opportunity to talk openly to affected family members are encouraged to take advantage of seeking out new information. Some women alter their perception of breast cancer risk, from one associated with breast cancer resulting in death to one in which an emerging belief about the possibilities for survival occurs."* (Esplen et al. 1998, pg 378)

Only one study has been reported that has developed an intervention based specifically on the SRM to challenge illness representations. Petrie et al. (2000) designed an intervention to alter representations of myocardial infarction (MI) in recently admitted patients. The intervention was designed to alter negative or 'catastrophic' beliefs concerning the consequences; timeline and control/cure of MI. Illness perceptions were assessed at baseline, prior to discharge from hospital and at 3 and 6 month follow up. Patients in the intervention group reported a greater understanding of their heart condition, were more ready to leave hospital and reported greater intention to attend rehabilitation than a control group receiving standard care. Analysis at follow up also revealed that the intervention group were likely to have returned to work faster than controls.

Although specific details of the intervention were not provided it is likely that the intervention involved cognitive behavioural strategies to alter illness representations. Cognitive Behaviour Therapy (CBT) fits well with the SRM as it is based on the premise that psychological problems are related to *'faulty patterns of thinking and*

behaviour' (Enright 1997). The cognitive aspect of treatment is therefore aimed at identifying and raising awareness of patterns of thought and the underlying assumptions, examining evidence on which the thoughts are based, challenging negative thoughts and learning coping strategies (Enright 1997). The efficacy of cognitive behavioural therapy has been demonstrated across a range of conditions including depression, generalised anxiety disorder, hypochondiasis and psychological problems in cancer patients (reviewed by Enright 1997). Recent studies have specifically applied cognitive behaviour therapy to improving mood and self esteem in breast cancer patients (Edelman and Kidman 2000, Edelman et al. 1999). It is possible that these techniques may be applied to help women at increased risk of breast cancer who are having psychological problems adjusting to their risk. CBT could be used in women at increased risk of breast cancer to raise awareness of cognitive and emotional representations of breast cancer, restructure unrealistic negative representations of the disease (e.g. beliefs about treatment), gain a realistic degree of control and improve coping strategies. However such interventions raise ethical dilemmas. Breast cancer *is* a serious and life threatening disease and to date there are few proven mechanisms by which to control or reduce risk. Negative beliefs about breast cancer may therefore be more realistic and attempting to challenge these beliefs may be maladaptive (Taylor et al. 1991, Affleck et al. 1987). Enright (1997) warns that care must always be taken when adapting cognitive behavioural therapy to any new area:

> *" Without this increased understanding of what works for whom and why, we should remain cautious of overenthusiastic claims for efficacy and of the clumsy application of genetic cognitive behavioural theory being made to fit increasingly diverse disorders"* (Enright 1997, pg 1815).

11.5.2 Psychological issues in detection behaviour

The SRM proposes that perceptual and conceptual representations are linked. It may be hypothesised therefore that individuals who are provided with a label for being 'at increased risk' of developing breast cancer may start searching for symptoms to match this label. Previous research has found that individuals holding strong

cognitive and emotional representations are more likely to seek medical care for undiagnosed symptoms (Cameron et al. 1993). It is possible therefore that both the strong illness representations derived from experiences of breast cancer and the label of being 'at risk' may encourage regular BSE. However these factors may also lead to preoccupation with breast cancer, excessive BSE and health anxiety.

> *"I think there is a tendency to imagine at the very beginning, when you know that you know there is a possibility of getting breast cancer, I think there is a tendency to imagine that you do have lumps and that you're forever testing and examining yourself"* (Appleton 1999, quote from telephone focus group study, personal communication).

A number of studies have indicated that women with a family history of breast cancer engage in excessive BSE. Approximately a third of women with a family history of breast cancer report that they practise BSE more than once a month (Lerman et al. 1994a, Brain et al. 1999). Epstein et al. (1997) reported that 8% of a sample of over 1000 women with a first degree relative with breast cancer practised BSE excessively (at least once a day). Although increased BSE has been associated with a number of factors such as ethnicity, perceived risk and intrusive thoughts about breast cancer, few explanation have been provided for this behaviour (Lerman et al. 1994a, Epstein et al. 1997). Lerman et al. (1994a) suggest that the behaviour may reflect obsessive-compulsive disorder or a hypochondrial response to breast cancer in the family. However in a discussion of hypochondria, Costa and McCrae (1985) warned that taking a psychiatric view to classifying individuals into pathological groups can often hide what is infact a continuous variable. They suggest that examining the phenomena from a psychological standpoint might provide a clearer explanation that applies across a range of individuals. The concept of health anxiety represents a continuum of orientation towards health concerns and could be applied to this issue. The development and maintenance of health anxiety has been explained from a cognitive behavioural perspective (Salkovskis and Warwick 1986, Warwick and Salkovskis 1990). Cognitive variables (i.e. beliefs about symptoms) derived from past experience or information are activated following certain incidences (i.e. experience of symptoms) and result in anxiety over health. Health anxiety is associated with attentional bias and misinterpretation of stimuli and can lead to either avoidance or excessive reassurance seeking. The theoretical

explanation of health anxiety overlaps with the concepts and processes of the SRM. However, the SRM also provides a clear link between cognition and coping strategies that has been omitted from the cognitive behavioural theory of health anxiety (Hadjistavropoulos et al. 1998). Both of these models can help explain avoidance or excessive BSE in women at increased risk of breast cancer. For example, from the SRM perspective, avoidance or excessive BSE in women at risk of breast cancer may be conceptualised as a misguided coping mechanism arising from particular cognitive and emotional representations stimulated by awareness of risk and experiences in the family.

It is likely that breast cancer related symptoms might also activate emotional and cognitive representations of the disease. Cameron et al. (1998) reported a trial of tamoxifen in women with breast cancer in remission. Hormonal symptoms associated with tamoxifen use were found to increase worries about breast cancer and BSE in comparison to a placebo-controlled sample. Cameron et al. (1998) suggested that the side effects of tamoxifen triggered representations of breast cancer and coping mechanisms including BSE. This holds implications for women at increased risk of breast cancer who maybe experiencing hormonal symptoms (eg menstrual problems, the menopause or symptoms resulting from chemoprevention). Additional information concerning the symptoms of breast cancer and the nature of hormonal symptoms in each case may help reduce levels of anxiety in each of these groups. Easterling and Leventhal (1989) also suggest that even non-cancer ('neutral') symptoms may induce distress. In a study of breast cancer survivors the experience of neutral symptoms (eg tiredness, pain) were associated with heightened cancer worry. The authors suggest that neutral symptoms may act as cues to ones mortality and vulnerability that can then activate threat cognitions. Provision of support from the familial breast cancer clinic and availability of urgent appointments for suspicious symptoms may be instrumental in alleviating anxiety about BSE and detecting breast cancer. Appleton (2000) found that women attending the familial breast cancer clinic in Edinburgh gained reassurance and security from the clinic. They felt closely monitored and privileged to be able to receive specialist care not available to other women.

11.5.3 Response to diagnosis

Applying the SRM to this population may also hold implications in the scenario of a diagnosis of breast cancer. Women holding different representations of the disease are likely to respond to diagnosis in different ways. For example, beliefs concerning the control of breast cancer may be associated with psychological adjustment to diagnosis. However, there has been limited research addressing women's response to developing breast cancer following information about their risk status. In a retrospective study, Petrisek et al. (2000) assessed the experience of breast cancer diagnosis in individuals with and without a family history of the disease. Women with a family history of the disease were less likely to delay symptom presentation, more likely to consult with specialists, felt comfortable about their treatment decision and more likely to have received adjuvant therapy than those without a family history of the disease. The authors noted that previous experiences of family members and representations of breast cancer appeared to be influence response to diagnosis. Petrisek et al. (2000) found that patients with a family history were more likely to be influenced by the experience of others and to fear recurrence when making treatment decisions.

The authors conclude that

> *" Physicians...should check that patients perceptions regarding morbidity and mortality are in proper perspective. Considering the importance placed on the opinions of family members and the similar experiences of others it may be beneficial to include relatives in treatment discussions".* (Petrisek et al. 2000, pg 141).

If women are helped and supported during diagnosis, treatment and recovery it is possible that these experiences will be less anxiety provoking for the next generation in the family.

11.5.4 Illness perceptions of health professionals

Buick (1997) found that health professionals held different representations of breast cancer when compared to healthy individuals. Although the study reported in this thesis was not designed to assess beliefs of health professionals, women in the general population sample who reported to have experience of breast cancer at work,

showed different perceptions of breast cancer to those without professional experience of the disease. A number of researchers interested in illness representations have suggested that inconsistent views between patients and health professionals may invoke problems in quality of care, particularly when the health care provider does not reach beyond the medical view of the illness (Buick 1997, Heijmans et al. 2001). Heijmans et al. (2001) systematically investigated the incongruence between the views of GPs and their patients suffering from a chronic illness (diabetes or osteoarthritis). Inconsistencies between illness perceptions held by patients and their GP were demonstrated for both diseases and the level of incongruence was associated with poorer physical and mental health. Further analysis also revealed that incongruence predicted health status and health care use even after patient perceptions were controlled. The different pattern of results between the patient groups led the authors to suggest that incongruence has a larger impact in disease with less clear treatment strategies. These results suggest that the perceptions of breast cancer held by health care professionals involved with genetic services may influence women's response to their risk status. GPs, consultants and genetic counsellors need to be aware of their own beliefs concerning breast cancer and breast cancer risk, potential conflict with perceptions and experiences of the counselee and the impact of this on the provision and comprehension of cancer risk information.

11.6 DIRECTIONS FOR FUTURE RESEARCH

This thesis reports the first studies that have directly used the SRM to address variation in response to risk of familial breast cancer. Although the results are promising there are a number of additional research questions that need to be addressed before interventions based on this approach can be designed and implemented.

As discussed in a previous section (11.3.3c) the validity of measures to assess components of the SRM require further research and additional dimensions relevant to individuals at increased risk of disease compared to patient populations need to be explored. These include beliefs regarding personal control over risk, efficacy of screening and detection methods as well medical prevention of the disease in

question. There is also a need to examine women's beliefs about genetics and inheritance and how they interact with representations of disease (Marteau and Senior 1997). Qualitative research is required to explore these perceptions in a systematic manner in order to fully appreciate representations of at risk populations.

Subsequent studies are also required to test the causal nature of the model. The SRM is dynamic and includes sets of interrelated factors making causality difficult to unravel. In addition, the factors involved are difficult to manipulate in an experimentally controlled manner. However a number of other designs have been discussed that may provide insight to causal mechanisms. These includes the use of longitudinal prospective designs to examine the impact of changing experiences and illness perceptions on levels of distress, intervention based studies and also studies designed in order to investigate the confounding impact of factors such as trait negative affectivity.

The research in this thesis was designed to assess a particular section of the SRM and did not include an assessment of coping. Although coping strategies form a major component of the model it was not feasible to measure coping given the large number of measures already included in the questionnaire. A number of studies have investigated a range of coping strategies in patient populations (Hagger and Orbell 2001). However, associations with illness representations or outcome are often elusive and tests of the mediation of illness representations to outcome via coping strategies have also proved unsuccessful (Scharloo et al. 1998, Heijmans 1998). It is possible that the use of general coping measures may not be sensitive enough to detect effects and it is therefore necessary to develop specific measures of coping responses relevant to the population in question.

Research assessing coping in women at increased risk of breast cancer has tended to focus on coping style rather than coping strategies. A monitoring coping style has been associated with increased distress in samples of women at increased risk of breast cancer and ovarian cancer (Lerman et al. 1996, Schwartz et al. 1995). However the implications for these results on intervention design are unclear (Lerman et al. 1996). Although qualitative studies have investigated how women

cope with an increased risk of breast cancer no measures are currently available (Appleton et al. 2000). Further research is required to investigate and assess specific coping strategies used within this population as well as satisfaction with these strategies. It is likely that cognitive and emotional representations of breast cancer will evoke different coping strategies and there is a need to examine the associations between coping, illness representations and adaptive or maladaptive adjustment to risk.

Another component of the SRM that has been neglected to date is the construct of appraisal. There are no studies to date that have explicitly investigating how individuals appraise their coping strategies and the process by which individuals alter their representations or coping behaviour. Research into this dynamic process is methodologically difficult and qualitative studies may be most informative in providing an insight. An understanding of how individuals appraise and change components of the SRM may provide information regarding potential effective intervention techniques.

Previously in this chapter the influence of trait negative affectivity on illness representation was discussed (see 11.3.2). A number of other personality factors may also influence illness representations including trait optimism. This construct has been associated with psychological adjustment to diagnosis and treatment in breast cancer patients and may also influence representations of women at increased risk of breast cancer (Carver et al. 1994). Optimists are likely to hold more positive representations, for example, concerning the duration and control of illness. Preliminary evidence suggests that beliefs concerning the prevention of breast cancer are associated with trait optimism in women at increased risk of breast or ovarian cancer (Audrain et al. 1997). Research into the SRM needs to incorporate and control for dispositional factors whilst maintaining a focus on specific components of the model that can be targeted in interventions. As well as personality factors a number of other beliefs may also influence illness representations for example beliefs about genetics and inheritance. These factors also need to be considered to gain a full understanding of the development and impact of illness representations in individuals facing genetic predisposition to cancer.

Leventhal et al. (1997) suggests that there may be an overlap between representations of disease and representations of self. The impact of risk on representations of self needs to be addressed in these populations. Nerenz and Leventhal (1983) discuss the integration between illness and self-concept. They identified three types of relations between illness and self-system in patient samples. Firstly, perceiving illness as permeating all aspects of life ('total'), secondly, perceiving only a portion of life to be effected by the illness ('encapsulated') or thirdly, perceiving oneself as healthy but at risk of acute disease episodes ('risk'). Kemp et al. (1999) assessed illness representations, aspects of self-schema in relation to illness, coping and adjustment in epilepsy patients. Ability to contain the effects of the illness was associated with avoidant coping and patients who perceived their illness as having pervasive effects on their lives showed heightened levels of distress. Different relations between breast cancer risk and self-concept were identified in a telephone focus group study of women attending the familial breast cancer clinic in Edinburgh (Appleton et al. 1999). Two examples highlighting different impacts of breast cancer risk on self-concept are given in the quotes below.

> *"I think you never know what's going to happen in the future and I wouldn't, I'm not going to spend my time worrying about something that might not happen or might not happen for a long time"*

> *"I think maybe the hardest thing I've had to do is accept that, that this will be an ongoing fear there will never come a time in my life when I will know, at least with current medicine as it is, there probably will never come a time when I will think, well this is something I won't get".* (Appleton, 1999, quote from telephone focus group study, personal communication).

Nerenz and Leventhal (1983) suggest that the experience of emotion such as depression in response to health threats may drive the illness to penetrate ones self-system. It is possible that awareness of a genetic predisposition to breast cancer, representations of the disease and emotional response to risk may impact of women's self-concept leading to further adjustment problems. The association between these factors requires further investigation in this population.

This thesis has demonstrated that illness representations influence women's emotional adaptation to risk. Section 11.5.2 also discussed how the SRM could enhance our understanding psychological issues regarding detection behaviour. Illness representations are also likely to play an important role in women's decisions about risk management and behavioural outcomes including participating in screening, clinical trials or prophylactic surgery. For example, the uptake of prophylactic surgery has been associated with heightened risk perception (Stefanek et al. 1995, Hatcher et al. 2001). However, beliefs regarding the efficacy of screening, consequences and treatment control of breast cancer may be predictive constructs. It is important to have insight into the decision making process in order that women can be helped to make the decision that is best for them and to ensure that decisions are made on up to date knowledge about breast cancer and not on unrealistic representations or misconceptions of the disease.

The work reported in this thesis may also hold implications for future work on psychoneuroimmunological interactions involved in breast cancer risk. Negative emotional states including anxiety and depression have been shown to have immunosuppressive effects (Selye 1976). Research has attempted to demonstrate the clinical implications of these findings for oncology by examining the impact of psychosocial factors on onset, progression and mortality from cancer including the impact of personality and stress on breast cancer onset and prognosis (Giraldi et al. 1997, Butow et al. 2001, Maunsell et al. 2001). Results in this area have however been inconsistent because of methodological difficulties (Edelman and Kidman 1997, Jensen 1991, Fox 1995). Research has tended to use retrospective, cross-sectional designs, lack control for other risk factors and use small sample size with insufficient statistical analysis (Jensen 1991). A few studies have addressed the impact of psychosocial interventions on survival time in metastatic breast cancer although conflicting results have been found (Spiegel et al. 1989, Edelman et al. 1999). Given possible associations between psychosocial factors and susceptibility to breast cancer it is feasible that emotional response to risk, illness representations and coping may influence phenotypic expression of risk in individual carrying a genetic predisposition. Research in this area would need to incorporate genetic components (including penetrance rates of different mutations etc) into the vast array of factors

already involved in psychoneuroimmunological reactions. Although a fascinating area for research the reactions are likely to be extremely complex and difficult to examine. This research would require multi-professional expertise and collaboration.

11.7 CONCLUSION

A theoretical approach was utilised within this thesis in order to address clinical research issues. This study was the first to apply the SRM to healthy women at increased risk of breast cancer in order to understand possible mechanisms by which experiences of breast cancer in the family influence psychological adjustment to breast cancer risk. The research reported in this thesis has presented a number of psychosocial factors that account for variability in distress in women at increased risk of breast cancer and discuss how these factors might interrelate. This research has identified a number of methodological issues with using the SRM in this population including the need to develop sound measures; explore the contribution of additional constructs pertinent to individuals with a genetic predisposition to cancer (eg perceived preventability, susceptibility); examine conceptual overlap between concepts and investigate issues of causality. Despite these limitations the SRM has been a useful framework with which to explore emotional response to risk and has potential to guide further research into psychological response to risk including the decision making process and behavioural outcomes.

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APPENDICES

Appendix I
Published paper: G.Rees, A.Fry and A.Cull (2000)'A family history of breast cancer: women's experiences from a theoretical perspective.' *Social Science and Medicine*. 52, 1433-1440.

Appendix II
Copies of the questionnaires

Appendix III
Additional analysis

PERGAMON

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A family history of breast cancer: women's experiences from a theoretical perspective

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Abstract

Individuals at increased risk of developing breast cancer due to their family history of the disease face a number of uncertainties. Personal cancer risk estimates are imprecise and current methods for early detection or prevention are not 100% effective. It is therefore not surprising that adverse psychosocial outcomes have been described within this population. Research attempting to predict the incidence of distress and dysfunction in individuals at increased risk of cancer has been largely a-theoretical and has overlooked a number of potentially important predictive variables. In particular, the influence of personal experience of cancer through involvement with affected relatives has been neglected. There are strong theoretical grounds for hypothesising that dimensions of personal experience may influence response to cancer risk. This paper discusses the potential impact of personal experience on risk perception, illness representations and decision-making. Systematic research in this area may improve predictions of outcome of cancer genetic counselling and inform the clinical process.

Keywords: Cancer; Psychosocial issues; Personal experience; Illness perception; Genetic risk.

Introduction

Breast Cancer is the most common form of cancer in women (Cancer Research Campaign, 1999). A small proportion of cases (about 5%) are caused by a germline genetic mutation which predisposes an individual to developing breast cancer, often at an early age (Evans et al., 1994). Women with a family history of breast cancer may be at risk of inheriting such a mutation and, relative to the general population, are therefore at increased risk of developing this disease.

Increased public awareness about the genetic basis of breast cancer has led to a growing number of familial cancer clinics which offer individuals genetic counselling about their risk and advice about risk management. Risk estimates are derived from family history details including the number of relatives who have suffered from breast cancer and the age at which they were diagnosed. A minority of women may be offered genetic testing to confirm they are at high risk. The growing demand for these services requires that resources such as mammography and clinical examination be targeted to women at greatest risk.

Psychosocial studies have been concerned with the uptake and outcome of these genetic services. A significant minority of women attending genetic counselling show high levels of distress, intrusive worries about breast cancer and misperceptions of risk which are not modified by genetic counselling (Lerman et al., 1995; Lloyd et al., 1996; Hopwood et al., 1998; Cull et al., 1999). There are concerns that inaccurate risk perceptions and distress may interfere with recommended health care actions for women at increased risk and provoke inappropriate behaviour in those at low risk (Lerman & Schwarz, 1993; Kash, Holland, Halper & Miller, 1992; Epstein et al., 1997). Associations between these variables are not fully understood and we are currently unable to predict which individuals will develop high levels of distress or hold misperceptions of risk.

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During genetic counselling the objective details of the family history are elicited. The personal experience of this family history is not likely to be explored in detail. Personal experience is shaped by exposure to and involvement in the physical care of ill relatives, as well as the emotional and social consequences of breast cancer in the family. It has been frequently documented that the experiences of relatives of cancer patients are extremely stressful (Northouse, 1995). This may be more so if the observing relative is aware of their own risk of developing the disease.

The subjective experience of breast cancer in the family may help to explain variation in women's response to breast cancer risk. However, research on this construct has been limited. This paper argues there are strong theoretical reasons for believing that dimensions of that experience are important mediators of risk perception and response to genetic risk information. This paper discusses the potential impact of experience from a theoretical perspective, in order to provide hypotheses to direct future research. The paper is divided into three main sections. The first section deals with bias in risk perception. Examples of cognitive bias in risk perceptions of relatives of cancer patients are outlined and discussed with reference to experience of cancer in the family. This is followed by a discussion on bias particular to genetic risk. The second section looks at how illness representations may be influenced by illness in the family and how representations may be altered by personal genetic risk information. The third section examines the impact of experiences on decisions and behaviour and ends with a discussion on how women's emotional response to their experiences may influence the decision-making process.

Risk perception and bias

Since the aim of genetic counselling is to provide the individual with risk information, the recall and accuracy of risk perception pre- and post-counselling have received much attention. Inaccurate perceptions of risk are commonly held and persist after genetic counselling (Lloyd et al., 1996; Cull et al., 1999). Inaccurate perceptions of risk, whether overestimated or underestimated, could have detrimental effects on psychological well-being and adherence to screening.

A number of factors including the method of presenting risk information have been shown to influence risk perception (Hallowell & Richards, 1997). Tversky and Kahneman (1974) described three heuristics that cause bias in risk perceptions when people reason using uncertain information such as probabilities. The influence of these heuristics has been demonstrated within the genetic counselling situation (Shiloh, 1994) and may be linked to experiences of cancer in the family.

Availability

Easily Recalled events are judged as more probable. Events that are more salient, familiar, recent and imaginable are perceived as more likely. This implies that exposure to a disease in family, friends or through the media has the potential to influence risk perceptions. Indeed the risk of health threats such as breast cancer and AIDS which receive much media attention are often overestimated (van der Pligt, 1998). Experience of breast cancer in family, friends and work colleagues has also been associated with heightened risk perceptions (Helzlsouer, Ford, Hayward, Midzenski & Perry, 1994, Drossaert, Boer & Seysel, 1996). Wardle (1995) found that women at increased risk of ovarian cancer and women in the general population showed positive associations between personal risk estimates and the number of friends and family who had died of cancer. We would predict that women at increased risk of breast cancer who have had closer and more frequent contact with relatives who have suffered from breast cancer, would be expected to show increased risk perceptions. Women who have recently suffered a diagnosis or bereavement in their family might also show elevated perceptions of risk.

Representativeness

Information about similarity and stereotypes are used to make judgements. Individuals are inclined to place emphasis on perceived similarities, but fail to consider the reliability of this evidence. Individuals often neglect the impact of probabilities or the effect of sample size on the representativeness of that observation. For instance, parents often refer to the degree of parental resemblance when judging their child's risk of developing an adult onset genetic disorder, such as Huntingtons disease (Shiloh, 1994). The degree to which a woman feels she resembles relatives in her family who have suffered from breast cancer may influence her perception of risk.

> I felt that I would get breast cancer as my body was similar to my mother's in many ways — she had fibroids and a hysterectomy — she had gall stones and had her gall bladder removed. I had both these operations by my late thirties. (J. Zatz, 1996, p. 28)

We may predict that women who feel they resemble relatives who have suffered from breast cancer, either physically, demographically or in personality, may have heightened perceptions of their risk. Indeed, studies have shown that individuals' feelings of susceptibility to familial cancer increase as they near the age at which their relative was diagnosed (Richards, Hallowell, Green, Murton & Statham, 1995; Brain et al., 2000).

Anchoring and adjustment

Individuals are biased towards a preconceived idea about their level of risk when provided with new risk information. The initial risk estimate is said to act as an 'anchor', which is adjusted following risk information. Shiloh and Saxe (1989) found that perception of risk after genetic counselling for congenital disorders was strongly influenced by expectations before counselling.

Studies have shown that individuals often make categorical judgments of risk and reduce risk to binary form, such as 'dangerous or safe' (Redelmeier, Rozin & Kahneman, 1993; Shiloh, 1996). A minority of individuals at increased risk of breast cancer believe that development of cancer is inevitable (Cull et al., 1999). This belief is likely to form a strong anchor that influences the way in which subsequent risk information is interpreted.

There is little information about what factors influence individuals' estimates of their risk status. It is likely that experiences in the family contribute to preconceptions of personal risk. Shiloh and Saxe (1989) found that experience of genetic disorder within the social circle was associated with higher risk expectations. It is possible that the proportion of family members affected by breast cancer may contribute to women's expectation of their level of risk.

> I think it's inevitable because there's no female members of my family who haven't had it.... It's scary. (Appleton, 1999, quote from telephone focus group study, personal communication).

Genetic risk and bias

Understanding of familial disease risk is further complicated by misconceptions about genetics. Lay concepts of inheritance are often based on resemblance and the joint inheritance of multiple physical and personality traits (Davison, 1996). There is a tendency to believe that susceptibility to illness is associated with similar personality and physical dimensions (Richards & Ponder, 1996). These beliefs often persist even after scientific, Mendelian accounts of inheritance have been provided (Richards, 1996). Beliefs about the inheritance pattern in the family, based on the relations and resemblance to affected relatives, may have a strong impact on perception of risk.

> When I was young my mother attributed her own breast cancer diagnosis to birth order. She talked about being the affected first born daughter of an affected first born daughter of an affected first born daughter. She told me that as a first born daughter in this line, I should expect to encounter the disease as well. With the diagnosis of one of my mother's younger sisters when I was 25, my mother stopped talking about the disease as a problem for first born daughters. Instead she dwelt on the personality traits that her affected sister shared with their mother — a certain intensity and vulnerability to stress looming large among them. (E. Macke, 1996 p. 32).

It is also widely believed that a greater proportion of inheritance is acquired from the same sex parent (Richards & Ponder, 1996). Indeed women with a paternal family history of breast cancer are underrepresented at genetic clinics (McAllister, Evans, Ormiston & Daly, 1998). It has been suggested that women with a paternal family history have lower perceptions of risk because of limited understanding of how a predominately female disorder can be passed on by males (Green, Richards, Murton, Statham & Hallowell, 1997).

Illness representations

Perceptions of breast cancer and beliefs about the disease are likely to influence how an individual reacts (in terms of thoughts, feelings and decisions) to their own risk status. A theoretical perspective that takes such beliefs into account is the study of lay beliefs and representations of illness (Leventhal Meyer & Nerenz, 1980).

Illness representations refer to peoples' perceptions of and beliefs about an illness. Leventhal's (1980) Self Regulatory Model states that reactions to health threats are mediated by these representations. Illness representations may develop from a variety of sources including direct experience of illness and medical care, experience of illness through family, friends and the media, as well as ideas inherent in cultural beliefs and language. Generally five dimensions of illness representations have been distinguished: identity of the threat (its symptoms and label); cause (e.g. infection, genetic, stress); time line (duration and development); consequences (including somatic and psychosocial); controllability in terms of prevention and cure (Leventhal & Benyamini, 1997).

Illness representations may differ widely depending on experience and culture and are dynamic and changing. Variation in representations will lead to different responses to the same health threat. Illness representations have been associated with psychological, behavioural and medical responses to a range of chronic illnesses (Scharloo & Kaptein, 1997). For example, Buick (1997) found that breast cancer patients who reported high levels of distress and functional disruption were those who perceived the disease as long in duration, with severe consequences, and who held strong

beliefs of self blame and diminished belief in the possibility of cure or control.

Genetic risk and illness representations

The experiences of women with a family history of breast cancer are likely to have a strong impact on their representations of the disease, particularly beliefs about the consequences, cure and control of breast cancer. Women with the same objective family history may have had differing levels of exposure to the effects of breast cancer and have witnessed different consequences of the disease. Experiences may range from exposure to positive role models, who survived breast cancer and coped well with the disease, to more negative experiences in which relatives suffered physically and mentally before dying.

> I'm lucky in the fact... that I have... more surviving relatives that have had breast cancer and are fine and that's a big inspiration... You think well its beatable its not a death sentence (Appleton, 1999, quote from telephone focus group study, personal communication).

These experiences and the subsequent representations derived from them are likely to have a profound effect on women's responses to genetic risk information including their emotional adjustment and screening behaviour. For example, Payne (1990) found that women's beliefs about the cause and control of breast cancer mediate the decision to practice breast self-examination. Many of these women were found to have experiences of breast cancer in friends and family, but the degree to which these experiences determined illness representations was not investigated.

We have little understanding of how awareness of genetic predisposition to breast cancer will affect representations of the disease. Being informed about genetic risk is most likely to influence beliefs about the cause of breast cancer but may also alter beliefs about its controllability and cure. Senior, Marteau and Weinman (2000) reported an experimental study in which participants were provided with hypothetical test results for either heart disease or arthritis. Participants who were informed that the test was genetic were more likely to attribute the illness to genes and less to lifestyle, and to rate both conditions as less preventable, than participants who were not informed of the nature of the test.

However, this study was conducted on participants with limited experience of the illness. It remains to be seen how knowledge about genetic risk will influence the beliefs of clinical populations with a family history of the disease in question. It is possible that the consequences of the illness witnessed in the family may influence beliefs about both breast cancer and genetic illness.

Decisions and behaviour

Women at increased risk of breast cancer are faced with a number of difficult decisions about risk management for example, whether to attend for screening, perform breast self-examination, participate in chemoprevention trials, have genetic testing or prophylactic surgery. Efficacy of these measures is not known. Given the lack of clear advice on the best course of action, women may be strongly influenced by their own experiences of breast cancer in their family and also by the decisions of other family members at increased risk. It is important to understand this decision-making process in order to help individuals interpret risk information and reach the decision that is best for them.

Early studies of human reasoning and decision-making attempted to discern processes that individuals use in order to make optimal decisions. Based on the utility theory individuals were thought to make decisions by assessing the probability and utility (importance) of events in order to maximise positive outcome (von Neumann & Morgenstern, 1947). Evidence for this rational decision-making strategy has remained controversial (Neumann & Polister, 1992). It is now accepted that such decisions are based upon a *subjective* interpretation of both probabilities and utilities. The theory was therefore generalised to the subjective expected utility theory (SEU) which proposes that optimal decisions are based upon the decision maker's personal expected utility of various outcomes.

Wroe, Salkovskis and Rimes (1998) assessed subjective reasons for undergoing predictive testing in two settings. The first used hypothetical scenarios to elicit reasons for genetic testing in student volunteers. The second examined reasons given by individuals who had contemplated genetic testing for a variety of disorders. The ratio of personal reasons for and against testing predicted the decisions made. In support of the SEU theory the prediction was enhanced when the reasons were weighted according to their relevance. Beliefs about the pros and cons of screening and preventative measures are likely to be influenced by a number of factors including other people's experience with the disease or behaviour. For example, Wroe et al. (1998) provided a scenario in which a woman refused mammography since she believed it to be a cause of breast cancer, based on experiences of family and friends.

Health behaviour models

Theories aimed at understanding health-related behaviour have been developed from the general decision-making perspective. A number of models have been developed based on the premise that individuals make a

rational analysis of the costs and benefits of possible behaviours. Two of these models will be discussed here. The best known model of health-related behaviour is the health belief model (HBM). This was initially designed to explain and predict compliance with preventative behaviours such as screening and immunisations (Rosenstock, 1966; Becker, 1974). According to this model readiness to engage in health behaviour depends on four beliefs: perceived susceptibility to the health threat; perceived seriousness of the health threat; perceived benefits of action; perceived costs of the behaviour. An individual will be most likely to engage in preventative behaviour if they regard themselves as susceptible to a serious illness and consider some preventive behaviour to have more benefits than costs. Once an individual is ready to act, behaviour is triggered by cues (Rosenstock, 1966). Cues may be internal, such as bodily states, or extraneous (e.g. health messages in the media).

Components of the model, including perceived susceptibility and barriers to action, have predicted breast self-examination among women in the general population (Champion, 1987). The HBM has been used with limited success to explain poor adherence to screening in women at increased risk of breast cancer (Kash et al., 1992). Relative to controls, women with a family history of breast cancer show higher levels of perceived susceptibility and perceive breast cancer as more severe (Wellisch, Gritz, Schain, Wang & Siau, 1991; Drossaert et al., 1996). However, the impact of a family history of breast cancer on screening behaviour remains unclear. Studies have reported both positive associations between a family history of breast cancer and screening behaviour (Lerman, Rimer, Trock, Balshem & Engstron, 1990) and no difference in breast self-examination and mammography attendance between women with and without a family history (Wellisch et al., 1991; Alagna, Morokoff & Bevett, 1987).

It may be that the subjective experiences within the family rather than a family history per se are important in forming the component beliefs of the HBM. In the studies of daughters of breast cancer patients, Wellisch, Gritz, Schain, Wang and Siau (1992) and Wellisch, Schains, Gritz and Wang (1996) examined daughters' experiences and perceptions of their mothers' illness. A number of aspects of experience appeared important determinants of adjustment. These included the daughter's age and degree of involvement with her mother when suffering from breast cancer and whether the mother had died (Wellisch et al., 1992). The daughter's perceptions of changes in mother's quality of life, daily living and self-image in terms of attractiveness and sexuality also appeared important factors (Wellisch et al 1996). These experiences may not only contribute to the perception of the severity of the disease but also act as cues to impel the individual to seek information about their own risk. Loss of relatives to breast cancer as well as recent diagnosis of breast cancer in family members has been shown to prompt women to attend breast cancer familial clinics (Richards et al., 1995). How often daughters of breast cancer patients talk to their mother about breast cancer has been positively associated with frequency of performing breast self-examination (Benedict, Coon, Hoomani & Holder, 1997).

Women with the same objective family history may have developed different beliefs about their personal susceptibility to breast cancer, the severity of the disease and its consequences, depending on their experiences. Some women may be more exposed to cues about their risk, depending on breast-cancer-related events and communication styles in the family.

A more general theory of behavioural decisions that has been applied to health-related behaviours is the theory of reasoned action (TRA) (Ajzen & Fishbein, 1980). This model suggests that intentions to perform a particular behaviour arise from both personal attitudes towards the behaviour and also social influence. Attitudes towards the behaviour may be positive or negative depending on the perceived consequences of the behaviour. Social influence, known as 'subjective norm' refers to the perceived expectations of important others. The theory was later extended to the theory of planned behaviour (TPB) with the addition of 'perceived behavioural control' in order to help explain behaviour that is not entirely under volitional control (Ajzen, 1991). The model proposes that intentions to perform a behaviour will be strong if an individual holds a positive attitude towards the behaviour, perceives him/herself to have control over the behaviour and believes that significant others expect him/her to perform it.

The theory of reasoned action/planned behaviour has rarely been applied to issues raised by cancer genetics. Devellis et al. (1990) assessed the predictive value of both the TRA and TPB on colorectal cancer screening for individuals at increased risk and controls. In both groups, intention to participate in screening was significantly associated with attitude toward the test. The predictive power was increased when perceived behavioural control was included in the model. Subjective norm only predicted intention to participate in screening in the control group. The authors suggest that subjective norm was not predictive in the high-risk sample due to its low variability. The majority of individuals in this sample reported that important others wanted them to undergo screening.

Whilst this model has not been explicitly tested in women at risk of breast cancer, studies suggest that relatives may actively encourage or dismiss utilisation of genetic services. For example, daughters of breast cancer patients are often encouraged to perform breast self-examination by their mothers (Benedict et al., 1997) and a minority of women attend familial breast cancer clinics

following advice from another family member (Brain et al., 2000). The beliefs of family members and communication styles within the family may influence both attitudes towards the behaviour as well as providing strong subjective norms.

Decision-making and emotions

Distress has been found to be an important barrier to screening programmes in women at increased risk of breast cancer (Lerman & Schwarz, 1993). Negative associations have also been found between levels of anxiety and frequency of performing breast self-examination (Kash et al., 1992). Distress and anxiety may also interfere with the decision-making process (Mann 1992). However, we have limited understanding of why some women show high levels of distress whilst others do not. It is likely that women's experiences of breast cancer in their family contribute to levels of anxiety and distress. For instance, women who have suffered bereavement due to breast cancer, who are particularly close to affected relatives or who have relatives currently undergoing treatment may show higher levels of distress. Zakowski et al. (1997) found that women at increased risk of breast cancer who had a parent die from cancer showed significantly higher levels of intrusive thoughts about cancer than women who had not suffered such bereavement.

Choices about risk-management strategies such as prophylactic surgery may be made in order to reduce anxiety. Stefanek, Helzlsouer, Wilcox and Houn (1995) found that women who opted for prophylactic surgery reported more worries about breast cancer than those who had decided against or were not considering surgery. Given that these women all had similar objective family histories, it is possible that the subjective experiences in the family may have contributed to breast cancer worry.

Given the serious nature of the decisions women face it is important to understand women's emotional response to breast cancer events in the family and how this may affect the decision-making process. Wroe et al (1998) found that emotions were considered as pros and cons when contemplating genetic testing. Both initial emotional reactions to genetic testing and emotions anticipated after testing appeared important.

> I am too anxious not to have the test" "I will get upset and anxious if I get a bad result (Wroe et al., 1998, p. 618).

Anticipated emotions are commonly cited when considering genetic testing. Lerman et al. (1995) found the main reason against genetic testing for breast cancer was concern about emotional reactions. Women anticipated anxiety, depression and impaired quality of life following a positive test result. Anticipating future emotions is extremely difficult and individuals tend only to consider emotions immediately following the decision rather than how they will feel in the longer term (Redelmeier et al., 1993). Women's experiences of breast cancer in their family and their beliefs about the disease are likely to have a large impact on how they anticipate their response to a positive test result.

Conclusions

Individuals at increased risk of breast cancer face much uncertainty about if and when cancer will develop and decisions about how to manage this risk. To date we are unable to predict which women will have difficulties adjusting to their genetic risk.

This review has shown that there are strong practical and theoretical reasons for believing that women's experiences of breast cancer in their family will influence how they think and feel about their own risk. Two women with the same objective risk of developing breast cancer may have had completely different experiences of breast cancer in their family. It is these experiences which may invoke different responses to genetic risk information. For example women may have had different levels of exposure to the disease, witnessed different medical, somatic and psychosocial consequences of the illness and identify with ill relatives to varying degrees.

This paper has shown theoretically how these experiences may influence women's perceptions of their risk, representations of breast cancer and decision-making processes. Ultimately these experiences may influence women's emotional and behavioural response to risk. We are now in a position to design hypothesis-driven studies to test the predictions suggested throughout this paper.

We need to understand the role of women's experiences of breast cancer in their family in how women conceptualise cancer risk, interpret and cope with risk information, and make subsequent decisions. Risk counselling in future may be more effective in helping women achieve an accurate perception of their risk and reducing adverse psychological consequences if it begins by taking account of the counsellees' pre-existing framework of beliefs based on their personal experience.

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Appendix II

INFORMATION SHEET
Personal views about breast cancer.

What is this study about?
This study is part of a 3 year research project funded by Imperial Cancer Research Fund to investigate women's beliefs about breast cancer. Many women feel anxious about the risk of breast cancer. This anxiety may be associated with experiences of the disease in family and friends. It is important to understand women's experiences and personal views about breast cancer in order to provide appropriate support. In this study we are interested in the beliefs of women in the general population and of women with a family history of breast cancer who attend the Ardmillan Familial Breast Cancer Clinic. We are interested in comparing the beliefs of women with and without a family history of breast cancer and to explore the associations between experiences of breast cancer, beliefs about breast cancer and levels of distress.

What is the purpose of this research?
The Ardmillan Familial Breast Cancer Clinic is a research led clinic where women with a family history of breast cancer are counselled about their risk and offered regular screening.
We have found that a small number of women who attend the clinic suffer from high levels of anxiety about their risk of breast cancer. The ultimate aim of this research project is to understand how women's experiences and beliefs about breast cancer are associated with this anxiety. Understanding the cause of such anxieties will help us to provide the best possible support to these women and to target psychological support to those who need it.

What is involved for me if I take part?
If you would like to take part in this study you will need to sign and return the enclosed consent form. We will then send you a questionnaire pack which we will ask you to complete and return. This will contain a number of questions about your background (marital status, number of children etc.), your experiences of breast cancer in your family, your personal views about breast cancer, your worries and concerns about breast cancer and your day-to-day feelings. We must stress that there are no right or wrong answers to any of the questions asked, and we are most interested in your own personal views.

What if I do not wish to take part?
If you do not wish to take part please let us know by returning the consent form having ticked the appropriate box. We can assure you that if you do not wish to take part in this study this will in no way affect any of the services you receive now or in the future. You are free to withdraw from the study at any time if you decide you do not want to complete the questionnaire.

What happens to the information I provide?
All information collected as part of this study will be treated as **highly confidential**. Only research staff involved in this project shall have direct access to the information collected. Information from the study may be passed to GPs and clinicians at the Ardmillan Familial Breast Cancer Clinic.

Can I find out more about the study?
If you would like to discuss this study further with someone who is not directly involved please contact: *Joyce Campbell, Genetic Research Nurse, Breast Unit, Western General Hospital, Crewe Road, Edinburgh, EH4 2XU. Tel: 0131 537 1615.* Joyce helped run the Ardmillan Familial Breast Cancer Clinic for a number of years but is not directly involved with this research project.

Principal Researcher: Gwyneth Rees, Department of Clinical Psychology, Outpatients Building, First Floor, Western General Hospital, Crewe Road, Edinburgh, EH4 2XU.
Tel: 0131 5371838 Email G.Rees@icrf.icnet.uk

Pilot experience questionnaire

You have already been asked a lot of questions for the family history form, but this does not tell us what your own experience of breast cancer within your family has been like. It is impossible to take into account the whole range of experiences people have had, but these questions aim to help us to understand how different experiences affect what people think, feel and do about their own risk.

Please complete the following 2 questions by giving the number which applies (or circle "Don't know")

How many of your relatives who have had breast cancer were personally known to you? _____

How many, if any of these relatives have died because of breast cancer? _____

At this stage we don't want to burden you with too many questions about your experience of breast cancer within your family and we realize that some of the things we are asking may be upsetting for you. We would like to get an idea of what dealing with breast cancer in your family has meant for you. For the following few questions please think about ONE RELATIVE who has or is suffering from breast cancer whose illness has particularly affected you. We realise that experience varies throughout the illness but we would like you to think of your lasting impression of their illness.

What relation was this one person to you (eg mother, sister etc)? _____

How close were you?	Not at all close	Quite close	Very close
	1	2	3

What age were **they** when breast cancer was first diagnosed? _____

What age were **you** when their breast cancer was first diagnosed? _____

How are they now?	Alive and well	Alive but unwell	Died from this cancer	Die from other causes
	1	2	3	4

22b. If this person died how long ago was that? _____

	(please circle the relevant response)			
	Not at all	A little	Quite a lot	Very much
Did their illness/treatment cause them much physical suffering?	1	2	3	4
Did their illness treatment cause them much emotional distress?	1	2	3	4
Overall, how upsetting was their illness for you?	1	2	3	4
Overall how well do you think they coped with their illness/treatment?	1	2	3	4
Do you think you are like them in general?	1	2	3	4
Do you feel that your relationship with this person changed when they developed cancer?	1	2	3	4
Do you feel that your role in the family has changed because of your experiences of breast cancer?	1	2	3	4
How much do you feel your life plans have changed because of the risk of cancer in your family?	1	2	3	4
Did you talk about your relative's cancer with the rest of your family at the time?	1	2	3	4
Do you talk now about your own risk with your family?	1	2	3	4
Do you talk now about your own risk with your family?	1	2	3	4

Experience questionnaire for the increased risk sample (Sample A)

Personal Experience of Breast Cancer

You have already been asked a lot of questions about your family history, but this does not tell us what your own experience of breast cancer within your family has been like. It is impossible to take into account the whole range of experiences people have had, but these questions aim to help us understand how different experiences affect what people think, feel and do about their own risk.

15. How many of your relatives who have had breast cancer were personally known to you? _____

16a. Have any of these relatives been diagnosed with breast cancer recently (ie within the last 5 years). Yes [ ] No [ ]

16b. If yes, please could you let us know how long ago these relatives were diagnosed?

_____

17a. How many, if any of these relatives have died because of breast cancer? _____

17b. Please could you let us know how long ago these relatives died?

_____

*We would like to get an idea of what dealing with breast cancer in your family has meant for you. For the following few questions please think about **ONE RELATIVE** who has or is suffering from breast cancer whose illness has particularly affected you. We realise that experience varies throughout the illness but we would like you to think of your lasting impression of their illness.*

18. What relation is this person to you (eg mother, sister etc)? _____

19.	How close were you?	Not at all close	Quite close	Very close	Extremely close
		1	2	3	4

20. What age were they when breast cancer was first diagnosed? _____

21. What age were you when their breast cancer was first diagnosed? _____

22a.	How are they now?	Alive and well	Alive but unwell	Died from this cancer	Die from other causes
		1	2	3	4

22b. If this person died how long ago was that? _____

		Not at all	A little	Quite a lot	Very much	Extremely
23.	Did their illness/treatment cause them much physical suffering?	1	2	3	4	5
24.	Did their illness treatment cause them much emotional distress?	1	2	3	4	5
25.	Overall, how upsetting was their illness for you?	1	2	3	4	5
26.	Overall how well do you think they coped with their illness/treatment?	1	2	3	4	5
27.	Did this person hold a positive attitude towards their illness?	1	2	3	4	5
28.	Do you think you are like them in body shape and size?	1	2	3	4	5
29.	Do you think you are like them in personality?	1	2	3	4	5
30.	Do you feel that your relationship with this person deteriorated when they developed cancer?	1	2	3	4	5
31.	Do you feel that your role in the family has changed because of your experiences of breast cancer?	1	2	3	4	5
32.	How much do you feel your life plans have changed because of the risk of cancer in your family?	1	2	3	4	5
33.	Do you feel that your experiences have brought the family closer together?	1	2	3	4	5
34.	To what extent have your experiences been positive?	1	2	3	4	5

Experience items for the general population sample (Sample B&C)

Personal Experience of Breast Cancer

We would like to ask you about any family members or friends who have suffered from breast cancer.

15a. Have any of your relatives ever suffered from breast cancer?

Yes ☐ No ☐

If no, please go to ***question 16.***

15b. Please list what relatives have suffered from breast cancer (eg mother, sister etc)

_____ _____ _____ _____

16. Have any of your family **or close friends** suffered from breast cancer **recently**? (ie in the past 12 months)

Yes ☐ No ☐

17. Does your work bring you into contact with cancer patients on a regular basis?

Yes ☐ No ☐

If yes, please give details

_____

_____

_____

18. Do you have any other experiences of breast cancer you would like to tell us about?

_____

_____

_____

_____

Experience items for the follow-up sample

Since you last filled in this questionnaire about 3 months ago have you attended the Ardmillan Familial Breast Cancer Clinic for an annual check up?

Yes ☐

No ☐

Since you last filled in this questionnaire, can you think of any experiences that may have changed your thoughts and feelings about breast cancer?

Yes ☐

No ☐

If yes, please let us know about these experiences by ticking the box (s) and describing your experiences. *(Please continue on a separate sheet of paper if necessary)*

☐ Events at the clinic _____

_____

_____

☐ Family experiences _____

_____

_____

☐ Experiences of friends _____

_____

_____

☐ Experiences at work _____

_____

_____

☐ Media Reports _____

_____

_____

☐ Other _____

_____

_____

Illness Perceptions Questionnaire- Revised (50 item) (IPQ-R50). Adapted to assess perceptions of breast caner in healthy women.

Item classification.
The subscales are colour coded for east of interpretation in this Appendix. Items that are reversed scored or removed in the IPQ-R38 are also highlighted.

Key:

Identity
Timeline acute/chronic
Timeline cyclical
Consequences
Treatment control
Personal control.
Illness coherence
Emotional representations
Causal items

* Item reversed scores
- Items removed from IPQ-R38.

Your views about breast cancer

We would now like to ask you some questions about your personal views about breast cancer. There are no right or wrong answers and we are most interested in your personal opinion.

Listed below are a number of symptoms that you may or may not associate with breast cancer. Please indicate by ticking in the space given, if you believe the symptom is related to breast cancer.

		This symptom is related to breast cancer.
a.	Hard or tender growths in body	.....
b.	Soreness in body	.....
c.	Skin changes	.....
d.	Pain	.....
e.	Sore Throat	.....
f.	Nausea	.....
g.	Breathlessness	.....
h.	Weight Loss	.....
i.	Fatigue	.....
j.	Stiff Joints	.....
k.	Sore Eyes	.....
l.	Wheeziness	.....
m.	Headaches	.....
n.	Upset Stomach	.....
o.	Sleep Difficulties	.....
p.	Dizziness	.....
q.	Loss of Strength	.....

We are interested in your own personal views about breast cancer.

Please indicate how much you agree or disagree with the following statements about breast cancer by ticking the appropriate box.

Item	VIEWS ABOUT BREAST CANCER	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
1	There is a lot patients can do to control symptoms of breast cancer					
2	Breast cancer has a negative impact on patients -					
3	Symptoms of breast cancer come and go in cycles					
4	Breast cancer is very unpredictable					
5	Symptoms of breast cancer are beyond patients' control * -					
6	Breast cancer makes me feel afraid					
7	Patients experience breast cancer symptoms pretty much all of the time *-					
8	Breast cancer is a mystery to me					
9	Breast cancer strongly affects the way patients see themselves as people -					
10	Breast cancer doesn't make any sense to me					
11	Breast cancer lasts for a long time					
12	The negative effects of breast cancer can be prevented (avoided) by treatment					
13	The symptoms of breast cancer change a great deal from day to day					
14	Recovery from breast cancer is largely dependent on chance or fate * -					
15	Breast cancer strongly affects the way others see patients					
16	I have a clear picture or understanding of breast cancer *					

	VIEWS ABOUT BREAST CANCER	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
17	Breast cancer has serious financial consequences					
18	Breast cancer causes difficulties for those who are close to patients					
19	Symptoms of breast cancer will be around whatever patients do * -					
20	Breast cancer lasts a short time *					
21	Treatment can control breast cancer					
22	What patients do can determine whether breast cancer gets better or worse					
23	I worry a lot about breast cancer -					
24	Breast cancer is likely to be permanent rather than temporary					
25	I get depressed when I think about breast cancer					
26	Breast cancer is easy to live with * -					
27	Breast cancer makes me feel anxious					
28	Nothing patients do will affect breast cancer *					
29	Patients have the power to influence breast cancer					
30	Breast cancer does not have much effect on patients' lives *					
31	Breast cancer is present all the time					
32	The symptoms of breast cancer are puzzling to me					
33	Breast cancer does not worry me *					
34	Breast cancer goes through cycles in which it gets better and worse					
35	Breast cancer improves in time [a1]					

[a1] Item number 35 is in Treatment control subscale in IPQ-R50. In the IPQ-R38 item 35 is moved to Timeline acute/chronic subscale where it is reversed scored.

	VIEWS ABOUT BREAST CANCER	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
36	When I think about breast cancer I get upset					
37	Breast cancer lasts for a lifetime					
38	Treatment is effective in curing breast cancer					
39	There is very little that can be done to improve breast cancer *					
40	Breast cancer makes me feel angry					
41	Breast cancer is not a problem for patients * -					
42	Patients' actions will have no effect on the outcome of breast cancer *					
43	Breast cancer passes quickly *					
44	The course of breast cancer depends on the patient -					
45	The symptoms of breast cancer are distressing to me -					
46	Breast cancer has major consequences on patients' lives					
47	Breast cancer is a serious condition					
48	There is nothing which can help breast cancer patients*					
49	Breast cancer doesn't bother patients much * -					
50	I don't understand breast cancer					

Causes of breast cancer

We are interested in what <u>you</u> consider may be the cause of breast cancer. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that may cause breast cancer rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for breast cancer. Please indicate how much you agree or disagree that they are causes of breast cancer by ticking the appropriate box.

POSSIBLE CAUSES	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
Stress or worry					
Hereditary - it runs in the family					
A germ or virus					
Diet or eating habits					
Chance or bad luck					
Poor medical care in the past					
Pollution in the environment					
Patients own behaviour					
Patients mental attitude e.g. thinking about life negatively					
Family problems or worries causes breast cancer					
Overwork					
Emotional state e.g. feeling down, lonely, anxious, empty					
Ageing					
Alcohol					
Smoking					
Accident or injury					
Patient's personality					
Altered immunity					
Hormonal					

In the table below, please list in rank-order the three most important factors that you believe to cause breast cancer. You may use any of the items from the box above, or you may have additional ideas of your own.

The most important causes of breast cancer:

1. _____

2. _____

3. _____

General Health Questionnaire

HAVE YOU RECENTLY

been able to concentrate on whatever you're doing?	Better than usual	Same as as usual	Less than than usual	Much less than usual
lost much sleep over worry?	Not at all	No more than usual	Rather more than usual	Much more than usual
been having restless, disturbed nights?	Not at all	No more than usual	Rather more than usual	Much more than usual
been managing to keep yourself busy and occupied?	More so than usual	Same as usual	Rather less than usual	Much less than usual
been getting out of the house as much as usual?	More so than usual	Same as usual	Rather less than usual	Much less than usual
been managing as well as most people in your shoes?	Better than most	About the same	Rather less well	Much less well
felt on the whole you were doing things well?	Better than usual	About the same	Less well than usual	Much less well
been satisfied with the way you've carried out your task?	More satisfied	About same as usual	Less satisfied than usual	Much less satisfied
been able to feel warmth and affection for those near you?	Better than usual	About same as usual	Less well than usual	Much less well
been finding it easy to get on with other people?	Better than usual	About same as usual	Less well than usual	Much less well
spent much time chatting with people?	More time than usual	About same as usual	Less time than usual	Much less than usual
felt that you are playing a useful part in things?	More so than usual	Same as usual	Less useful than usual	Much less useful
felt capable of making decisions about things?	More so than usual	Same as usual	Less so than usual	Much less capable
felt constantly under strain?	Not at all	No more than usual	Rather more than usual	Much more than usual

HAVE YOU RECENTLY

felt you couldn't overcome your difficulties?	Not at all	No more than usual	Rather more than usual	Much more than usual
been finding life a struggle all the time?	Not at all	No more than usual	Rather more than usual	Much more than usual
been able to enjoy your normal day-to-day activities?	More so than usual	Same as usual	Less so than usual	Much less than usual
been taking things hard?	Not at all	No more than usual	Rather more than usual	Much more than usual
been getting scared or panicky for no good reason?	Not at all	No more than usual	Rather more than usual	Much more than usual
been able to face up to your problems?	More so than usual	Same as usual	Less able than usual	Much less able
found everything getting on top of you?	Not at all	No more than usual	Rather more than usual	Much more than usual
been feeling unhappy and depressed?	Not at all	No more than usual	Rather more than usual	Much more than usual
been losing confidence in yourself?	Not at all	No more than usual	Rather more than usual	Much more than usual
been thinking of yourself as a worthless person?	Not at all	No more than usual	Rather more than usual	Much more than usual
felt that life is entirely hopeless?	Not at all	No more than usual	Rather more than usual	Much more than usual
been feeling hopeful about your own future?	More so than usual	About same as usual	Less so than usual	Much less hopeful
been feeling reasonably happy, all things considered?	More so than usual	About same as usual	Less so than usual	Much less than usual
been feeling nervous and strung-up all the time?	Not at all	No more than usual	Rather more than usual	Much more than usual
felt that life isn't worth living?	Not at all	No more than usual	Rather more than usual	Much more than usual
found at times you couldn't do anything because your nerves were too bad?	Not at all	No more than usual	Rather more than usual	Much more than usual

Cancer worry scale

The following questions ask about any concerns you may have regarding breast cancer. For each question please tick one box to indicate your answer.

During the past month, how often have you thought about your own chances of developing cancer? Would you say ..... *(Please tick one box to indicate your answer)*

- Not at all or rarely ☐
- Sometimes ☐
- Often ☐
- Almost all of the time ☐

During the past month, have thoughts about your chances of getting cancer affected your mood? Would you say...

- Not at all or rarely ☐
- Sometimes ☐
- Often ☐
- Almost all of the time ☐

During the past month, have thoughts about your chances of getting cancer affected your ability to perform your daily activities? Would you say.....

- Not at all or rarely ☐
- Sometimes ☐
- Often ☐
- Almost all of the time ☐

How concerned are you about the possibility that you might get cancer someday? Would you say.....

- Not at all ☐
- Somewhat ☐
- Moderately ☐
- Very concerned ☐

How often do you worry about developing cancer? Would you say....

- Not at all ☐
- Occasionally ☐
- Frequently ☐
- Constantly ☐

How much of a problem is worrying about cancer to you? Would you say....

- Not at all ☐
- Somewhat ☐
- Definitely ☐
- Severe problem ☐

Impact of Event scale

We are interested in knowing how people think about their risk of breast cancer. Please circle the appropriate number to indicate how frequently these comments were true for you during the last 7 days.

If you have not thought about your risk of breast cancer in the last 7 days please tick this box and go on to the next page.

I have not thought about the risk of breast cancer. ☐

		Not at all	Rarely	Some-times	Often
a.	I thought about it when I didn't mean to	1	2	3	4
b.	I avoided letting myself get upset when I thought about it or was reminded of it	1	2	3	4
c.	I tried to remove it from memory	1	2	3	4
d.	I had trouble falling asleep or staying asleep, because of pictures or thoughts about it that came into my mind	1	2	3	4
e.	I had strong waves of feelings about it	1	2	3	4
f.	I had dreams about it	1	2	3	4
g.	I stayed away from reminders of it	1	2	3	4
h.	I felt as if it wasn't real	1	2	3	4
i.	I tried not to talk about it	1	2	3	4
j.	Pictures about it popped into my mind	1	2	3	4
k.	Other things keep making me think about it	1	2	3	4
l.	I was aware that I still had a lot of feelings about it, but I didn't deal with them	1	2	3	4
m.	I tried not to think about it	1	2	3	4
n.	Any reminder brought back feelings about it	1	2	3	4
o.	My feelings about it were sort of numb	1	2	3	4

Key for subscales: Intrusion Avoidance

Appendix III

Item-scale (corrected) correlations.
Bold represents the proposed scale for each item

Item	Scale	r	n	p
1. Did their illness/treatment cause them much physical suffering?	**Traumatic experience**	**.63**	**110**	**.000**
	Coping	-.13	111	.164
	Resemblance	.19	114	.046
	Change	.38	114	.000
	Positive experience	-.17	114	.075
2. Did their illness treatment cause them much emotional distress?	**Traumatic experience**	**.62**	**110**	**.000**
	Coping	-.33	109	.001
	Resemblance	.11	111	.27
	Change	.21	111	.029
	Positive experience	-.03	111	.75
3. Overall, how upsetting was their illness for you?	**Traumatic experience**	**.42**	**110**	**.000**
	Coping	-.005	110	.96
	Resemblance	.38	113	.000
	Change	.51	113	.000
	Positive experience	.05	114	.59
4. Overall how well do you think they coped with their illness/treatment?	Traumatic experience	-.24	110	.011
	Coping	**.57**	**111**	**.000**
	Resemblance	.24	113	.011
	Change	-.006	113	.95
	Positive experience	.35	113	.000
5. Did this person hold a positive attitude towards their illness?	Traumatic experience	-.11	108	.25
	Coping	**.57**	**111**	**.000**
	Resemblance	.18	111	.056
	Change	.04	111	.65
	Positive experience	.36	111	.000
6. Do you think you are like them in body shape and size?	Traumatic experience	.19	110	.042
	Coping	.27	111	.018
	Resemblance	**.52**	**114**	**.000**
	Change	.22	114	.005
	Positive experience	.26	114	.002
7. Do you think you are like them in personality?	Traumatic experience	.29	110	.002
	Coping	.16	111	.101
	Resemblance	**.52**	**114**	**.000**
	Change	.40	114	.000
	Positive experience	.35	114	.000
8. Do you feel that your relationship with this person deteriorated when they developed cancer?	Traumatic experience	.34	110	.000
	Coping	-.26	111	.007
	Resemblance	.016	114	.87
	Change	**.18**	**114**	**.059**
	Positive experience	-.15	114	.11
9. Do you feel that your role in the family has changed because of your experiences of breast cancer?	Traumatic experience	.39	110	.000
	Coping	.067	111	.49
	Resemblance	.30	114	.001
	Change	**.30**	**114**	**.001**
	Positive experience	.088	115	.35

10. How much do you feel your life plans have changed because of the risk of cancer in your family?	Traumatic experience Coping Resemblance **Change** Positive experience	.15 .14 .22 **.23** .02	110 111 114 **114** 115	.12 .14 .02 **.014** .88
11. Do you feel that your experiences have brought the family closer together?	Traumatic experience Coping Resemblance Change **Positive experience**	.086 .29 .36 .13 **.38**	110 111 114 114 **115**	.37 .002 .000 .16 **.000**
12. To what extent have your experiences been positive?	Traumatic experience Coping Resemblance Change **Positive experience**	-.23 .40 .19 -.09 **.38**	110 111 114 114 **115**	.014 .000 .045 .32 **.000**

Item deletion analysis of the IPQ-R

Cases in which the Cronbach's alpha for each of the subscales from the IPQ-R is improved with an item omitted from the scale. For each subscale from each version of the measure (IPQ-R50 and IPQ-R38) the item deleted and corresponding alpha coefficient are provided.

IPQ-R subscale	Increased risk 50 item		Increased risk 38 item		General population 50 items		General population 38 items	
	Item deleted	Alpha	Item deleted	Alpha	Item deleted	Alpha	Item deleted	Alpha
Identity	1 2 3 11	.67 .67 .67 .67	-	-	3	.78	-	-
Timeline acute/chronic	-	-	35	.61	43	.65	35	.61
Consequences	2	.71	15 17	.58 .58	-	-	-	-
Personal control	19	.82	44	.79	-	-	-	-
Treatment control	12 35	.74 .73	12	.76	35	.63	12	.65
Illness coherence	-	-	-	-	-	-	-	-
Timeline cyclical	31	.38	-	-	31	.33	4	.49
Emotional representations	-	-	-	-	40	.88	-	-

Further test-retest analysis of the IPQ-R.

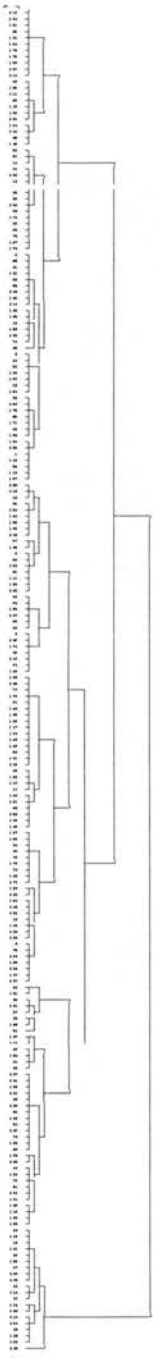
The Table below shows test re-test correlation coefficients for participants who did and did not report breast cancer related experiences between the first questionnaire and follow-up questionnaire.

Test re-test correlation coefficients for individuals with and without breast cancer related experiences between the questionnaires.

	IPQ-R50				IPQ-R38			
Subscale	**Participants with experience between questionnaires**		**Participants without experience between questionnaires**		**Participants with experience between questionnaires**		**Participants without experience between questionnaires**	
	N	**R**	**N**	**R**	**N**	**R**	**N**	**R**
Identity	32	.55**	42	.54***	-	-	-	-
Timeline Acute/chronic	30	.58**	37	.74***	29	.57**	37	.70***
Consequences	28	.73***	40	.80***	30	.76***	40	.70***
Personal control	29	.54**	39	.83***	29	.61***	39	.82***
Treatment control	28	.69***	40	.81***	29	.66***	40	.78***
Illness coherence	30	.68***	40	.69***	-	-	-	-
Timeline cyclical	30	-.20	40	.56***	30	.22	41	.56***
Emotional representations	26	.88***	38	.88***	26	.82***	38	.87***

***p<.001
**p<0.01

Dendrogram from hierarchical cluster analysis of IPQ-R subscales (Wards method) in the general population sample



Sobel test calculations for mediation models suggesting mediation.

Model	a	b	s_a	s_b	Test statistic	p value
11.1(ii)	.211	1.971	.098	.194	2.106	0.035
11.1(iii)	.173	1.069	.088	.402	1.581	0.11
11.4(iii)	-.0055	1.330	.002	.647	1.646	0.099
11.4(iv)	-.00067	8.441	.000	4.351	1.940	0.052
11.5	.926	-1.455	.434	.457	-1.77	0.076
11.6(iii)	.137	2.105	.061	.270	2.158	0.031
11.7(i)	.112	2.319	.029	.300	3.46	0.0005
11.8	.100	2.132	.048	.272	2.013	0.044